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Disclaimer: This report contains transcripts of interviews conducted in the course of the research and contains language that may offend some readers.

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Abstract

A framework to address key issues of neonatal service configuration in England: the NeoNet multimethods study

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Background: There is an inherent tension in neonatal services between the efficiency and specialised care that comes with centralisation and the provision of local services with associated ease of access and community benefits. This study builds on previous work in South West England to address these issues at a national scale.

Objectives: (1) To develop an analytical framework to address key issues of neonatal service configuration in England, (2) to investigate visualisation tools to facilitate the communication of findings to stakeholder groups and (3) to assess parental preferences in relation to service configuration alternatives.

Main outcome measures: The ability to meet nurse staffing guidelines, volumes of units, costs, mortality, number and distance of transfers, travel distances and travel times for parents.

Design: Descriptive statistics, location analysis, mathematical modelling, discrete event simulation and economic analysis were used. Qualitative methods were used to interview policy-makers and parents. A parent advisory group supported the study.

Setting: NHS neonatal services across England.

Data: Neonatal care data were sourced from the National Neonatal Research Database. Information on neonatal units was drawn from the National Neonatal Audit Programme. Geographic and demographic data were sourced from the Office for National Statistics. Travel time data were retrieved via a geographic information system. Birth data were sourced from Hospital Episode Statistics. Parental cost data were collected via a survey.

Results: Location analysis shows that to achieve 100% of births in units with ≥ 6000 births per year, the number of birth centres would need to be reduced from 161 to approximately 72, with more parents travelling > 30 minutes. The maximum number of neonatal intensive care units (NICUs) needed to achieve 100% of very low-birthweight infants attending high-volume units is 36 with existing NICUs, or 48 if

NICUs are located wherever there is currently a neonatal unit of any level. Simulation modelling further demonstrated the workforce implications of different configurations. Mortality modelling shows that the birth of very preterm infants in high-volume hospitals reduces mortality (a conservative estimate of a 1.2-percentage-point lower risk) relative to these births in other hospitals. It is currently not possible to estimate the impact of mortality for infants transferred into NICUs. Cost modelling shows that the mean length of stay following a birth in a high-volume hospital is 9 days longer and the mean cost is £5715 more than for a birth in another neonatal unit. In addition, the incremental cost per neonatal life saved is £460,887, which is comparable to other similar life-saving interventions. The analysis of parent costs identified unpaid leave entitlement, food, travel, accommodation, baby care and parking as key factors. The qualitative study suggested that central concerns were the health of the baby and mother, communication by medical teams and support for families.

Limitations: The following factors could not be modelled because of a paucity of data – morbidity outcomes, the impact of transfers and the maternity/neonatal service interface.

Conclusions: An evidence-based framework was developed to inform the configuration of neonatal services and model system performance from the perspectives of both service providers and parents.

Future work: To extend the modelling to encompass the interface between maternity and neonatal services.

Funding: The National Institute for Health Research Health Services and Delivery Research programme.

Contents

List of tables

List of figures

List of supplementary material

Report Supplementary Material 1 Literature review on neonatal mortality

Report Supplementary Material 2 Qualitative study information

Supplementary material can be found on the National Institute for Health Research Journals Library report project page (www.journalslibrary.nihr.ac.uk/programmes/hsdr/141908/#/documentation).

Supplementary material has been provided by the authors to support the report and any files provided at submission will have been seen by peer reviewers, but not extensively reviewed. Any supplementary material provided at a later stage in the process may not have been peer reviewed.

Glossary

BadgerNet Neonatal Electronic Patient Record The electronic patient record system used by the vast majority of neonatal units.

Birth centre A maternity unit usually staffed by midwives and often (but not always) obstetricians.

BLISS (Baby Life Support Systems) A UK charity working to provide the best possible care and support to all premature and ill babies and their families.

British Association of Perinatal Medicine An association of professionals who have a special interest in neonatal care. It was founded in 1976.

British Association of Perinatal Medicine standards A specification of nurse-to-infant ratios for staffing in neonatal care recommended by the British Association of Perinatal Medicine (see *Chapter 1*, *Clinical importance*).

Decision space The range of possible options; in this study, it refers to the range of possible configurations of unit locations.

Fitness function An objective function used to summarise in a single figure how close a solution is to achieving the set aims.

Healthcare Resource Group A standard grouping of clinically similar treatments used as a means of determining reimbursement for care services to NHS providers.

Heuristic method A practical method to provide a satisfactory solution to a problem without a guarantee of optimality.

Instrumental variable approach A method used in economic applications when a controlled experiment to test the existence of a causal relationship is not feasible and some correlation between the original explanatory variables and the error term is suspected.

Lower-layer super output area Geographic areas with approximately equal population sizes (a minimum population size of 1000 and an average population size of 1500).

Maptitude® (Caliper, Newton, MA, USA) A geographic information system used in this study for travel time estimation.

Non-dominated solution A solution is non-dominated if none of the objective or criterion functions can be improved in value without degrading some of the other objective values (see *Chapter 6*, *Dealing with multiple criteria: Pareto dominance*).

Nurse workload The number of nurses required to care for infants present in a unit (see *Chapter 1*, *Clinical importance*).

Objective space A set of objective or criterion values that are achievable from the decision space.

Pareto front A framework for partially evaluating a set of options with multidimensional outputs (see *Chapter 6*, *Dealing with multiple criteria: Pareto dominance*): set of non-dominated solutions.

Primipara A woman who has given birth to only one child.

Probit model A type of regression where the dependent variable can take only two values.

Royal College of Obstetricians and Gynaecologists A professional association, founded in 1926, with the objective to encourage the study and advancement of the science and practice of obstetrics and gynaecology.

SNUG (Supporting Neonatal Users and Graduates) An association providing mentoring and befriending services for parents of ill or premature babies in Devon.

Structural mean model A semiparametric model that uses instrumental variables to identify causal parameters.

Tobit cost model A statistical approach to regression that is used extensively in health economics.

Very low-birthweight infant An infant born weighing < 1500 g.

List of abbreviations

ANNP	advanced neonatal nurse practitioner
BAPM	British Association of Perinatal Medicine
BLISS	Baby Life Support Systems
CI	confidence interval
CV	coefficient of variation
DCE	discrete choice experiment
HES	Hospital Episode Statistics
HRG	Healthcare Resource Group
ICER	incremental cost-effectiveness ratio
ICU	intensive care unit
IMD	Index of Multiple Deprivation
IQR	interquartile range
IV	instrumental variable
LNU	local neonatal unit
LOS	length of stay
LPM	linear probability model
LSOA	lower-layer super output area
MOO	multiobjective optimisation
NDAU	Neonatal Data Analysis Unit
NICE	National Institute for Health and Care Excellence
NICU	neonatal intensive care unit
NNRD	National Neonatal Research Database
NSGA-II	Non-Sorting Genetic Algorithm II
ODN	Operational Delivery Network
OLS	ordinary least squares
OR	odds ratio
PPI	patient and public involvement
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
R&D	research and development
SCU	special care unit
SMM	structural mean model
SNUG	Supporting Neonatal Users and Graduates
SPEA	Strength Pareto Evolutionary Algorithm
VLBW	very low birthweight
WTE	whole-time equivalent

Plain English summary

When organising neonatal care in England, there is a tension between the centralisation and localisation of services. These services are currently provided at different levels, ranging from neonatal intensive care units (NICUs) to special care units. For the most-ill infants, intensive specialised care delivered in high-volume NICUs (i.e. those that see more ill infants) has been shown to deliver improved health outcomes; however, smaller local units provide easier access and reduce travel times for parents.

Geographic analysis and computer models were used to investigate a range of alternative scenarios for neonatal care, looking at the impact of greater and lower levels of centralisation. The models suggest that having fewer units, especially for intensive care, could potentially improve infant survival rates. Costs and resource implications (e.g. the number of nurses required), as well as the impact on parental travel time, were also investigated using these models.

The results demonstrate the advantages of different service configurations and provide a framework to assist policy-makers in planning neonatal services. We also looked at the most effective way to present these results using a range of graphical and visualisation tools.

When parents were interviewed, it was found that the health of the mother and the baby dominated their concerns. Parental overheads associated with entitlement to unpaid leave, food, travel, accommodation, baby care and parking were also revealed to be important concerns.

Further work is needed to investigate the relationship between maternity and neonatal services and to understand the impact of alternative configurations of care on infant health and well-being.

Scientific summary

Objectives

The aim of this study was to develop an analytical framework to address key issues in the configuration of neonatal services in England. The primary component objectives were to:

- Analyse neonatal service organisation and explore the trade-offs that are inherent in reconfiguration.
- Understand the benefits and costs, both to the NHS and to parents, of service centralisation and model the impact of different configurations. To use simulation, modelling and location analyses to understand the behaviour of this complex system and investigate trade-offs at the national level.
- Model costs and outcome changes associated with service reconfiguration. To explore the impact of service reconfiguration on clinical outcomes (e.g. mortality) and costs [e.g. neonatal bed-days, length of stay (LOS) and parent costs] and to undertake qualitative research on factors that families and policy-makers see as important in determining service configuration.
- Investigate the use of visualisation tools to communicate research findings. To understand the informational requirements of the key stakeholder groups and to research and develop effective communication tools to convey the research findings.
- Consult with the parents of neonatal infants. To ensure that the needs and concerns of parents and families are taken into account, to explore the best ways to communicate findings to parents and the public, and to involve them in decision-making in neonatal service configuration.

Methods

A wide range of approaches was employed. Descriptive statistics, location analysis, mathematical algorithms and simulation were used for modelling work. In the health economic analysis, cost and mortality models were developed and structured qualitative interviews with both policy-makers and parents were conducted. For data visualisation, user requirements and tools for communication were investigated, and patient and public involvement (PPI) workshops were held with parents.

Location analysis and modelling

Data sources

- The geographic areas used were 2011 lower-layer super output areas (LSOAs).
- Demographic data for LSOAs were obtained from the Office for National Statistics.
- Road travel times were obtained from a geographic information system (Maptitude® 2016; Caliper, Newton, MA, USA).
- Birth data (at the LSOA level) for 2013–15 were obtained from Hospital Episode Statistics.
- Neonatal unit designation was obtained from the 2015 National Neonatal Audit Programme report.
- Neonatal care data for 2013/14 were obtained from the 2015 National Neonatal Research Database (NNRD). Of the 161 neonatal units, 90% gave permission to access NNRD data [100% of neonatal intensive care units (NICUs), 85% of local neonatal units (LNUs) and 90% of special care units (SCUs)].

Location analysis

For both maternity and neonatal care configuration, there is a trade-off between competing objectives. Prioritising parent travel time entails more small neonatal units close to the locations of mothers' homes. Prioritising health outcomes entails fewer neonatal units providing intensive treatment and greater travel times.

When competing objectives exist, there are many possible solutions, and increasing performance in one objective is accompanied by a reduction in the performance of at least one other objective. This is known as the 'Pareto front'. We used a genetic algorithm to identify good solutions that approach the theoretical optimal Pareto front.

There are no national target times for access to maternity or neonatal care. We analysed mean and maximum travel times and the proportion of mothers within 30, 45 and 60 minutes of the place of care. The 30-minute target is most discriminatory between options and, therefore, it is the target that is most commonly referred to in this report.

The Royal College of Obstetricians and Gynaecologists recommends that obstetrician-led maternity units should deliver ≥ 6000 births per year to facilitate the 24/7 (24 hours a day, 7 days a week) presence of a consultant on site. Currently, only about 20% of births take place in units of this size. Our maternity location analysis shows that, to achieve 100% of births in units of this size, the number of birth centres would need to be significantly reduced: from 161 to approximately 70. However, with this reduction, the proportion of mothers attending units with ≥ 6000 births per year and travelling < 30 minutes would increase from 24% to 82% with reconfiguration (the maximum that is achievable based on our results). Although such large changes may be unrealistic, the proportion of mothers attending a unit with ≥ 6000 births per year within 30 minutes could be increased to approximately 70% with 120 units (with ≈90% of mothers living within 30 minutes of such a maternity unit).

The British Association of Perinatal Medicine (BAPM) recommends that all NICUs should admit ≥ 100 very low-birthweight (VLBW) infants per year. The genetic algorithm identified solutions in which all NICUs could meet this guideline and travel distances for parents could be reduced. This would, however, require moving the location of some NICUs. If NICUs are restricted to current NICU locations, meeting this BAPM guideline would require the closure of 12 of the current 48 NICUs, and this would increase travel times for parents.

Simulation

A discrete event simulation was used to further evaluate configurations. This mimicked admissions over time and modelled networks, transfers, different unit levels and capacities. The simulation placed infants in the closest appropriate unit (with sufficient capacity) and tracked infants, nurse workload in units, transfers and distance from mothers' home locations.

The simulation accurately predicted average travel times (within 2 minutes or within 10% of actual data). Predicted workload accuracy varied depending on proximity between units; for units that were ≥ 15 minutes apart, the typical error margin was ± 2–3 infants or ± 1 nurse-equivalent workload.

The simulation showed that the number of infants not cared for at their closest appropriate unit rises with capacity utilisations of > 60% of the maximum capacity, with a doubling of the number of infants who are > 30, 45 or 60 minutes from home when units run at an average of 75% of maximum capacity. The removal of network boundaries had a minimal impact on travel times.

The simulation also predicted that relative peaks in workload are significantly lower for higher-volume units. A unit with an average workload of 2–3 nurse equivalents will have a fourfold ratio of peak-to-trough nurse workloads (ratio of ninetieth-to-tenth percentile workloads), whereas a unit with average workload of 10 nurse equivalents will typically have a twofold ratio of peak-to-trough nurse workloads.

Two alternative scenarios, identified by the genetic algorithm, were tested in the simulation model. One scenario, selected to minimise travel distances while having all NICUs admitting ≥ 100 VLBW infants per year, reduced average travel times and increased the proportion of infants within 30, 45 and 60 minutes from home. In this scenario, the simulation replicated the expected benefits. The second scenario modelled significant centralisation of care (with 30 NICUs, 30 LNUs and 30 SCUs). In this scenario, travel times for parents were increased, but the number of nurses required to meet BAPM standards for 90% of the time was reduced by about 10%.

Economic modelling

The economic modelling explored the impact of service reconfiguration on clinical outcomes (e.g. mortality) and costs (e.g. neonatal bed-days, LOS and parent costs), and involved qualitative research on family and policy-maker preferences.

Mortality

Mortality for infants born at < 32 weeks of gestational age was estimated using semiparametric and parametric neonatal mortality models in both (1) high-volume units (≥ 100 VLBW admissions per year) and (2) NICUs. Sensitivity analysis accounted for potential bias attributable to imbalance in the distribution of extremely premature babies across treatment (hospital of birth) groups. Causal effect was estimated using an instrumental variable (IV) approach, using travel time or distance as an instrument. Secondary analysis of the relative effects of birth in a hospital with a NICU versus a SCU and versus a LNU used an IV approach with three instruments: travel time to the closest (1) NICU, (2) LNU and (3) SCU.

It was found that exposure to a high-volume unit at birth reduces mortality relative to birth in other neonatal units. A very preterm infant has a 5-percentage-point lower risk of death by being born in a hospital with a high-volume unit than being born in another neonatal unit (when travel time is used as the instrument) when semiparametric models are used and a 1.2-percentage-point lower risk when parametric models are used. Sensitivity analysis, excluding infants born at a gestational age of < 26 weeks, halves the mortality effect of birth in high-volume units compared with other units, but the estimates are imprecise. For babies born at a gestational age of < 32 weeks, being born in a hospital with a NICU appears to not affect the risk of death compared with being born in other units. The secondary analysis suggests that NICUs reduce mortality by 1.9 percentage points compared with LNUs.

Costs

The limitations of national Healthcare Resource Groups (HRGs) were investigated based on data analysis and interviews with policy-makers and the impact of high-volume units on LOS, and the costs of neonatal care for families were estimated.

Site visits and discussions with policy-makers made it clear that national neonatal HRG costs do not currently accurately reflect actual costs, mainly because units typically use average neonatal nursing costs across all infants and do not use the BAPM guidelines to attribute nursing to different levels of care. In addition, apportioning of costs (e.g. diagnostic costs) differs across neonatal units and other units within hospitals, and staff composition varies greatly; for example, junior doctors and advanced neonatal nurse practitioners cover similar tasks, but vary on the salary scale. Our evaluation focuses on (1) the impact of high-volume units on LOS and reimbursement and (2) costing reconfigurations based on nursing cost estimates from the model rather than relying on HRG costs.

The impact of service configuration on LOS was explored and LOS was costed using a microcosting approach. The analysis compared high-volume units with other units using an IV approach. LOS and costs were modelled using a log-normal distribution, whereas probit equations were used for SCU and LNU binary treatment indicators. The instruments and estimates (as for the mortality analysis) included covariates for gestational age, gestational age squared, birthweight, infant sex, last decile of the Index of Multiple Deprivation score, mode of delivery and number of fetuses.

The mean total LOS following a birth in a high-volume unit is 9 days longer and has a mean cost of £5715 more than for a birth in another neonatal unit. The mean total LOS following a birth in a LNU is shorter by 1–2 hospital days, and in a SCU it is shorter by 3–4 hospital days, relative to the mean LOS following birth in a NICU. The mean cost of a SCU birth is £1770 less than a birth in a NICU, although the effects are imprecisely estimated. In contrast, mean reimbursement costs for births in a LNU are £834 more costly than NICU births, but this result is not significant.

A linear regression cost model was developed to capture 'out-of-pocket' expenses that have an impact on family budgets. The Baby Life Support Systems (BLISS) data on parent costs suggested that key factors were entitlement to unpaid leave, food, travel, accommodation, baby care and parking. In addition, the support from the employer of the mother's partner can reduce costs, as can the availability of the partner to help.

What is important to families?

A flexible topic guide for individual interviews with parents was developed (after piloting prior to data collection). The semistructured probing questions were based on a review of the literature and PPI workshop feedback. From the transcripts of the interviews with parents, a thematic framework was developed to code relevant themes and subtheme factors.

The qualitative interviews found that people talked about the infant as a whole, rather than separating out risks of death and health problems. Families connected the health of the baby and the mother, considering the mother's health alongside that of the child. The interviews also highlighted other process outcomes (e.g. communication with the families and family support) and raised questions about the ability and willingness of parents to trade off health attributes with process attributes. Mothers were unlikely to want to sacrifice 'core' aspects of their baby's health for improvements in process outcomes.

Evaluation

We estimated the incremental cost-effectiveness based on a comparison of (1) high-volume units and all other units, and NICUs and other unit designations, and (2) three service reconfigurations from the simulation modelling.

For high-volume units compared with all other units, dividing the additional cost (£5715) by the reduction in neonatal mortality (absolute risk difference 0.012) results in a cost per neonatal life saved of £460,887. Costs and effects were discounted by 2.5% for the first 30 years of life, 3% from the 31st to the 75th year and 2.5% from the 76th to the 81st year (as recommended by the HM Treasury Green Book for longer term interventions[135]). This results in an incremental cost-effectiveness ratio (ICER) per life-year gained of £15,620. In addition, birth in a NICU is more clinically effective and cost saving, with an incremental cost per neonatal life saved of –£43,096 when compared with birth in a LNU.

Exploring three service configurations, it was found that nursing costs are the largest cost component, being approximately 18 times higher than travel costs and 33 times higher than transfer costs. Nursing costs also reduce during centralisation, because of economies of scale, and are the key driver for overall costs. Although centralisation increases family travel costs, it reduces costs from a societal perspective.

Data visualisation

Policy-makers, commissioners, clinicians and care workers, researchers, parents and the public all have an interest in the organisation of neonatal care. These diverse groupings generally have both different informational needs and expertise. Therefore, the way in which information is communicated, just as much as what is communicated, needs to reflect these differing requirements.

A range of potential formats and media for the presentation of findings from the study was investigated and a number of graphical methods and specific tools (some developed within our research) was identified to address audience requirements. Policy-makers emphasised the importance of clear methods to communicate technical outputs and their relation to strategic issues. Parents found maps, Pareto fronts and narrative- and picture-based presentations based on case study information powerful additions to, but not substitutes for, traditional text-based information. Importantly, any development of communication tools needs to start with a clear understanding of the objectives and the specific audience requirements.

Parental involvement

Five PPI workshops were carried out, which allowed the team to develop an ongoing dialogue with parents about the design of aspects of the project, the implications of the findings for neonatal services and parents, and how potential negative consequences of centralisation might be mitigated. Importantly, these workshops demonstrated that parents can be involved in complex, evidence-based discussions about the design and delivery of neonatal services if they are supported and provided with the relevant information in an accessible manner.

Conclusion

In this study, a structured approach is presented to address key questions in the configuration of NHS neonatal care services in England. Although many issues still need further investigation, it is believed that the research framework outlined here provides a valuable basis to support evidenced and informed policy-making in this area of health and care.

Funding

Funding for this study was provided by the Health Services and Delivery Research programme of the National Institute for Health Research.

Chapter 1 Clinical setting

Clinical importance

In England and Wales, 86,000 infants (around one in eight births) are admitted to neonatal units each year.[1]

Neonatal care comprises four levels of care, each with different staffing recommendations for nurse-to-infant ratios [summarised from the British Association of Perinatal Medicine (BAPM)]:[2]

1. Intensive care for the sickest infants involves care, such as mechanical respiratory support or the presence of an arterial line. The NHS service specification for neonatal critical care[3] requires all units to work towards a 1 : 1 nurse-to-infant staffing ratio in neonatal intensive care.
2. High-dependency care involves care, such as non-invasive respiratory support or parenteral nutrition. A 1 : 2 nurse-to-infant care ratio is recommended.
3. Special care may involve care, such as intranasal oxygen or nasogastric feeding. A 1 : 4 nurse-to-infant care ratio is recommended.
4. Transitional care is the lowest level of care, and may be shared between neonatal and maternity units. The mother is frequently considered the primary carer in transitional care, and no guidelines for staffing levels are given.

Neonatal units are frequently under significant pressure, and units are often required to transfer infants to another unit or work at higher than the BAPM guideline[2] for infant-to-staff ratios.[4] In 2007, the neonatal charity Baby Life Support Systems (BLISS) reported that:[5]

- Neonatal units were, on average, understaffed by over one-third.
- Over 6 months, neonatal units were closed to new admissions for an average of 24 days as a result of excessive demand.
- One in 10 units exceeded their capacity for intensive care for > 50 days during a 6-month period.
- Of neonatal units providing a full range of intensive care services, 65% did not have enough staffed cots for the infants admitted.
- One-quarter of twins or triplets were cared for in separate hospitals.

Reports other than those from BLISS have found units working above the BAPM guidelines on workload per nurse.[4] A unit's capacity may be limited either by the number or level of available cots or by the number of nurses.

The Department of Health and Social Care's neonatal planning toolkit[6] recommends an 80% occupancy of units, but the National Institute for Health and Care Excellence (NICE)[7] has reported that units often operate at higher occupancy levels. However, the ≈80% occupancy target often used in the NHS is frequently derived from whole hospital bed modelling,[8] and it may not be appropriate to extrapolate this to neonatal care.

There is some evidence to show that infant mortality increases when units work at higher occupancies.[9] General care is also compromised, with nursing activities more likely to be delayed or omitted when the unit is running at nursing ratios below the BAPM guidelines.[4]

In contrast to levels of care, neonatal units are described in terms of three basic levels[3,6] depending on the most complex care that can be provided. Neonatal intensive care units (NICUs) provide all levels of care, local neonatal units (LNUs) usually provide high-dependency care and special care units (SCUs) provide special care and a stabilisation facility prior to transfer to more specialist units. A further consideration is that care may be provided by a network of units[10] in which specialist care is centralised and low-level care is distributed across the network. The performance of one unit is then heavily dependent on the others, so that the reduction in capacity of one will increase workload in the others. This makes planning at a network level, rather than at a unit level, essential. In 2003, a Department of Health and Social Care report[11] highlighted the need for better tools for planning neonatal care.

The costs for parents for care that is distant from their home location is significant. In a recent report by BLISS,[12] the mean cost to parents of having an infant in neonatal care was £282 per week, or £2256 over the course of their child's stay.

The relationship between neonatal nursing levels and unit size on clinical outcome

Clinical outcome has been linked to the level of neonatal nursing provision. A 2013 systematic review of NICUs[13] concluded that lower nurse staffing was associated with poorer outcome. In an observational study,[14] a 10% decrease in the proportion of intensive care days in which one-to-one nursing was provided was associated with an increase in the in-hospital mortality rate of 0.6 deaths per 100 infants receiving neonatal intensive care per month compared with a median monthly mortality rate of 4.5 deaths per 100 infants per month.

In a study of 2585 very low-birthweight (VLBW) (< 1500 g) or preterm (gestational age of < 31 weeks) infants,[15] it was found that higher neonatal nurse staffing was associated with reduced mortality: at times when 1 : 1 staffing was achieved for intensive and high-dependency care infants, the risk of mortality was halved compared with at other times. Other outcomes may also be influenced by staffing levels, as increased neonatal nurse staffing has been associated with a reduction in the risk of bloodstream infection.[16]

In contrast, in a study of 850 moderately preterm infants (gestational age of 30–34 weeks),[17] higher nurse-to-infant ratios were not associated with improved outcome, although this may have been related to the later gestational age (infants with a later gestational age are often at lower risk of poor outcome, and generally require less intensive care).

Larger neonatal units have been linked to lower mortality. In a study of 48,237 VLBW infants born in Californian hospitals between 1991 and 2000,[18] there was a significant reduction in mortality associated with birth in higher-volume units. Compared with NICUs with ≥ 100 VLBW admissions per year, smaller units were associated with higher odds ratios (ORs) for death, ranging from 1.19 [95% confidence interval (CI) 1.04 to 1.37] to 2.72 (95% CI 2.37 to 3.12). The authors[18] suggest that 20% of deaths may have been preventable if intensive care was centralised in the higher-volume units.

In a large study of 20,554 infants,[19] birth in higher-volume units correlated with the survival of babies born at a gestational age of < 33 weeks. Infants who were born at < 33 weeks' gestation and admitted to a high-volume neonatal unit (defined as being in the upper quartile of care days) at the hospital of birth were at reduced odds of neonatal mortality (OR 0.70, 95% CI 0.53 to 0.92) and any in-hospital mortality (OR 0.68, 95% CI 0.54 to 0.85). The effect was most marked among infants born at < 27 weeks' gestation (OR 0.51, 95% CI 0.33 to 0.79).

It is noteworthy that in both of these large studies,[18,19] the analysis was restricted to infants born in hospitals with large NICUs. The analysis did not extend to infants who were transferred to such units. The interaction between place of birth and place of neonatal intensive care is complex. For infants who were born in 1992/3 with a birthweight of < 2000 g, the mortality OR was 2.4 times higher if the birth was in a hospital without a NICU, with the risk being lowest in the highest-volume regional intensive care units (ICUs).[20] Subsequent transfer to a regional NICU marginally decreased the disadvantage of birth at a hospital without a NICU. Taken alone, this might suggest that the advantage of a large NICU is conferred primarily on those infants born in hospitals with large NICUs. However, the analysis is confounded as infants who die before transfer are excluded, as are those who are not ill enough to warrant transfer. The advantage of delivery in, rather than transfer to, a hospital with a high-volume NICU remains uncertain.

In view of the possible link between volume and outcome, the BAPM[21] suggests that:

- Neonatal intensive care units in the UK should have a throughput of ≥ 100 VLBW infants per year (VLBW is defined as < 1500 g).
- Neonatal networks that include NICUs admitting < 50 VLBW infants should develop plans to amalgamate NICUs (or NICUs plus LNUs) to increase throughput.

Optimal size of birth centres

The Royal College of Obstetricians and Gynaecologists has drawn attention to the optimal size of obstetric units, based in part on requirements for consultant presence.[22] It has been suggested that the provision of a continuous consultant-led service should theoretically be possible if there is centralisation into units with ≥ 6000 births per year.[22]

The large majority of births (87%) in England take place in obstetric units, with 11% taking place in midwifery-led units and 2.4% taking place at home.[23] Although 11% of births are in midwife-led units, only 2% of all births in England take place in freestanding midwife-led units.[24] Obstetric-led units or alongside midwifery-led units (where the midwife-led unit is in the same site as an obstetric-led unit, with near-immediate access to specialist obstetric care if required) therefore account for 95% of all births. It is this very large majority of births that is the focus of the modelling study in this report.

To achieve more births in units with ≥ 6000 births per year requires an increase in the centralisation of service provision. The disadvantage is that it increases the travel distance for some mothers. There is no strong evidence in Featherstone *et al.*[25] or in a study of 3 million live births in France[26] that there is a link between travel time and outcome for VLBW infants. Nevertheless, data from term deliveries in the Netherlands suggest that mortality is correlated with the estimated car travel time from home.[27] Estimated car travel times of > 20 minutes were associated with an increased risk of mortality and adverse outcomes, and when travel time was used as a continuous determinant, the adjusted OR for mortality per minute increase of travel time was 1.01.[27] The authors noted that although the study attempted to adjust for confounding variables, it was possible that the results could be explained by variables other than travel time.

Likewise, in a study of 413,000 singleton births in Wales (1995–2009),[28] there was a positive correlation between time of travel to hospital and the adjusted risk of neonatal death. The correlation remained after allowance for various confounding variables (such as gestational age and social deprivation index), although the authors did not have information on type of onset of labour, or medical or surgical conditions affecting the mother or infant. Importantly, the Welsh study also looked at travel time to the closest maternity unit as opposed to the unit to which the woman was actually taken. They did not find any association with mortality, suggesting that the association between travel time and outcome may not be causal, but may reflect the medical condition of the mother or the infant, leading to lengthened travel to a tertiary centre. The overall conclusion was that there was no strong evidence of an association between mortality and the geographic location of maternity services.

As travel time increases, there is a risk of birth before arrival at hospital. In France, the rate of out-of-hospital births for mothers living > 30 km from their nearest maternity unit was double that for those living within 30 km of their nearest maternity unit.[29] Among primiparae (women who have borne only one child), out-of-hospital birth rates increased from 2.3 per 1000 for those living < 5 km from their nearest maternity unit to 7.5 per 1000 for those living ≥ 45 km from their nearest maternity unit. For women with a parity of four or more, the out-of-hospital birth rates were 5.4 per 1000 for those living < 5 km from their nearest maternity unit and 26.2 per 1000 for those living < 5 km from their nearest maternity unit. Although rare, there was an increase in deaths related to out-of-hospital birth, increasing from 4.0 per 100,000 births for distances of up to 14 km to 10.0 per 100,000 births for distances of ≥ 45 km or more, although death following out-of-hospital birth accounts for < 2% of all neonatal deaths.[26] These results suggest that, although there is no strong evidence for a link between distance and neonatal outcome, consideration should be given to avoiding excessive distances or travel times to avoid the low risk of out-of-hospital birth.

As there are no guidelines on targets for distance to maternity units in the UK, we examine a range of measures of access. These include mean and maximum travel times, and the proportion of mothers within 30, 45 and 60 minutes of a maternity unit.

Payment by Results and information on which this is based for neonatal care

Most neonatal units in England are reimbursed on their activity under the Payment by Results system.[30] In this payment system, commissioners pay health-care providers for each infant, taking into account the infant's health-care needs.[30] Healthcare Resource Groups (HRGs) are used to represent clinically similar treatments that use common levels of health-care resource. In neonatal care, there are five neonatal HRGs that are paid per occupied bed-day (Intensive Care XA01Z, High Dependency XA02Z, Special Care without external carer XA03Z, Special care with external carer XA04Z and Normal Care XA05Z), and a neonatal transport HRG is paid per patient journey (Neonatal Critical Care Transport XA06Z). Some units are still paid under the old 'block contract' system, and are assigned a fixed amount of money to deliver neonatal services. Typically, block contracts are now used only to help units maintain services and manage their finances in periods of severe financial pressures.

There is a per diem tariff assigned to each neonatal HRG. Units are paid for the number of occupied bed-days for each level of care and the number of critical care transport journeys, based on locally agreed tariffs assigned to the HRGs. The tariffs are based on information provided by units on the costs of providing these services, which they submit in the reference cost returns each year.[31] The NHS provides guidance on the allocation of costs for reference cost returns and this guidance has been modified over time to try to collect more accurate costing for neonatal services. However, there has been a significant delay in updating the data items that make up the HRG reference costs (units are currently still paid in accordance with the HRG 2001 data set when submitting cost information against revised HRG reference cost guidance)[32] and there is disparity in the way that trusts attribute costs between neonatal and paediatric services and how they apportion costs between the different neonatal HRGs. This delay has led to a 'price cost' gap, in which the costs of neonatal care may be different from the reimbursements received. For units paid on the Payment by Results system, there is a clear incentive to keep accurate records of costs for reference costs returns and the activity within the unit. For units on a block contract, there are fewer incentives, as their block contract is less explicitly dependent on the tariff system that the reference costs returns informs.

Chapter 2 Background

Across many health-care services, there is an ongoing tension between the expertise, efficiency and specialised care that comes with centralising resources and the provision of locally based services with their associated ease of access for users and community benefits. In neonatal care, this issue is further complicated by the organisation of care into regional networks, where different hospitals provide differing levels of care, and where capacity across, or even between, networks may be used when local capacity is exhausted. In addition, the interface between maternity and neonatal care in hospitals provides further complications and options for the organisation of services that affect service delivery.

This study builds on previous work conducted within the South West of England[33] to address many of these key issues. Modelling and geographic analysis are used to assess service distribution options; we also investigate the economic aspects of varying scenarios for service delivery. In addition, we assess the preferences of parents using these services.

The primary components of this study fall into the following areas:

- Sourcing and analysis of neonatal data – extensive descriptive statistical analysis was applied to data obtained from the National Neonatal Research Database (NNRD) held at the Neonatal Data Analysis Unit (NDAU), as well as Hospital Episode Statistics (HES).
- Geographic analysis – a range of mathematical methods were used to model service locations in order to analyse and optimise distribution of any number of units with the aims of meeting target admission numbers while reducing travel distances for parents.
- Simulation modelling – computer software was used to model alternative configurations of units or altered staffing levels. The model was used to predict system performance from the perspective of the service provider (e.g. average and peak loads, proportion of time when staffing meets BAPM guidelines,[2] and nurse requirements) and parents (travel distances and costs and the number of parents travelling more than a reasonable daily travel distance).
- Economic analysis – an exploration of the prediction for mortality and, if feasible, morbidity to service delivery statistics, and costs to both parents and service providers. We explore, through qualitative research and discrete choice experiments (DCEs), the factors that both families and policy-makers consider important in deciding how services should be organised and how these might be weighted when making decisions.
- Data visualisation – we consider key aspects of information presentation and communication of outcomes to a range of stakeholders in the context of the complexity inherent in many of the key findings.
- Parental involvement – through a series of workshops involving parents of neonatal infants, we have investigated the key factors of importance to users of neonatal services.

Chapter 3 Project aims and objectives

The central aim of our study was to provide an analytical framework that addresses many of the key questions relating to the configuration of neonatal services in England. It is hoped that this framework will then help inform policy and the development of new models of care in this area.

The main objectives of our study, as outlined in the proposal, are listed below:

- To analyse neonatal service organisation and investigate the trade-offs inherent in reconfiguration. The centralisation of services has benefits including increased throughput leading to increased expertise and a reduction in the spare capacity needed to deal with peaks in workload; however, centralisation may increase distances that parents need to travel. The effect is further complicated by the transition of the infant through different levels of care, and by the organisation of units into networks. Modelling and location analysis provide excellent tools for understanding the behaviour of this complex system. We use these tools to investigate that trade-off at a national level and address the following key questions.
    - How would reconfiguration (e.g. greater centralisation) affect unit throughput and parent travel times/distances?
    - What is the relationship between the number of units and the expected travel times and throughputs?
    - What is the average and maximum planned distance and travel time from parents' home location to the point of care? How does this vary across the country?
    - How does changing the number of on-duty staff affect the number of transfers and the travel distances for parents?
    - What happens to travel distances if network boundaries are removed?
    - How might conflicting objectives in service distribution be best balanced?
    - Given any fixed number of units, which locations would minimise travel times?
    - What is the expected impact of population changes?
- To model costs per infant and outcome change associated with service reconfiguration. Neonatal reference costs (e.g. HRGs) have limited utility for modelling as they assume a fixed infant cost regardless of the size of the unit and do not account for variation in nursing costs, which are dependent on configuration. We sought to model neonatal costs in significant detail in order to better predict the relationship between service configuration and costs. Having access to a recent BLISS survey on costs to parents, we also sought to better understand the relationship between network configuration and parental costs. We addressed the following key questions regarding this objective.
    - What components, and in what proportion, contribute to the costs of the different types of neonatal unit?
    - How would changes in the degree of centralisation of services affect the spare capacity needed to deal with peaks and troughs in workload? How would total costs be affected?
    - How would changes in the degree of centralisation of maternity (birth episode) and neonatal services affect parent travel distances and costs?
    - How does the degree of centralisation affect the requirement for local accommodation for parents?
- To investigate the use of visualisation tools to communicate our research outputs to stakeholder groups. A key aspect of dissemination in research is the use of effective tools and media to present information. In this context, it is also essential to recognise the different needs and expertise of the varying stakeholder groups. To this end, we assessed a number of different information visualisation representations in terms of these communication requirements. We addressed the following key questions.
    - What are the key stakeholder communities that need to understand our research outputs?
    - What are the information needs of the different stakeholder groups?
    - Which visualisation tools and media are best suited to convey information effectively to the range of identified users?

- To consult with the parents of neonatal infants to assess their preferences. It is essential to involve parents in the process of decision-making and priority-setting, and to ensure their representation in policy and debate about neonatal service organisation. Within our research, we engaged with parents through a series of workshops in which we elicited views and preferences about neonatal service delivery based on direct experience. These were complemented by qualitative interviews with parents conducted within the health economic research component of the project.

Chapter 4 Data sources

Geographic areas

All of the patient/parent location data used 2011 lower-layer super output areas (LSOAs). LSOAs are geographic areas with approximately equal population sizes (minimum of 1000, average of 1500). The home location of mothers was taken as the population-weighted centroid of each LSOA.[34]

Demographic data

The 2015 Index of Multiple Deprivation (IMD) for each LSOA was obtained from the Office for National Statistics.[35]

Travel time data

All travel times were based on estimated fastest road travel times. Travel times for patients and parents were taken from the postcode closest to the population-weighted centroid of the parents' LSOA. Travel times were estimated using Maptitude® 2016 (Caliper, Newton, MA, USA) with the MPMileCharter® (Winwaed Software Technology LLC, Wichita Falls, TX, USA) add-in.

Birth data

Births per LSOA for the 3 years from 2013 to 2015 were obtained from NHS HES[36] using Lightfoot Solutions® Signal-from-Noise tool version 2.1 (Lightfoot Solutions, Bracknell, UK). Births were defined as all admissions with a primary procedure code (Office of Population Census and Surveys) of R17–R27. Data obtained were aggregate numbers of births; no patient-level data were obtained.

Demographic projections

Demographic projections were obtained from the Office for National Statistics.[37]

Neonatal unit designation

Neonatal-unit-level designation was taken from the 2015 National Neonatal Audit Programme Report.[1]

Neonatal unit cost data

Neonatal unit costs were taken from the *NHS Reference Costs 2014–15*, published by the Department of Health and Social Care.[31] Neonatal costs are associated with critical care services and the HRG codes relative to neonatal care. The HRG system aims to define standard groups of clinically similar treatments that use common levels of health-care resource; it is used by the NHS to determine fair and equitable reimbursement for care services delivered by providers.

Predictions of NHS costs in the evaluation of simulations were based on the level of care received and a microcosting based on whole-time equivalent (WTE) nurses. Unit costs for WTE nurses are based on *Unit Costs of Health and Social Care 2014*.[38] The costs to families that we include in the model are travel time and vehicle operating costs. The unit costs applied to travel time are based on the Department for Transport's non-business costs of travel,[39] derived from a willingness-to-pay study of non-business travellers.[40]

Family cost data

Family costs were obtained from BLISS, a UK charity working to provide the best possible care and support for all premature and sick babies and their families. The data received are from a survey in which parents described the neonatal experience, the family condition and the expenses during the neonatal care period.

A total of 1347 of the questionnaires were returned; however, there was large amount of missing information on costs and family characteristics, which was attributable to the high number of questions and the large presence of free-text options.

Most of the data in the survey were incomplete and this can create some bias in family cost evaluation. Infant length of stay (LOS) data were missing in 6% of the overall observations, whereas the travel distance in miles from the parents' residence to hospital is missing for 29% of data, and the travel time from the parents' residence to hospital is missing in 81% of data. Other important data that were missing were regarding the age of parents (15%), the number of overnight stays in hospital and the relative cost (97%), the cost of childcare (90%), the costs of baby care (57%), the cost of food (32%), the cost of parking (48%), the cost of travel (23%) and the amount of unpaid leave (77%).

Neonatal data

Neonatal data were obtained from NNRD held by the NDAU hosted at Imperial College London.[41] These data originate from the 'BadgerNet' (Clevermed, Edinburgh, UK) neonatal electronic clinical care records kept in each unit.[42]

The following ethics and research and development (R&D) approvals were obtained:

- Integrated Research Application System – reference number 172210
- Research Ethics Committee – reference number 15/NW/0503
- Chelsea and Westminster Hospital R&D approval – reference number C&W16/022.

Out of 161 neonatal units, 145 (90%) gave permission to access NNRD data. Of all units, 41 out of 41 NICUs (100%) gave permission, 67 out of 79 LNUs (85%) gave permission and 37 out of 41 SCUs (90%) gave permission.

Data were obtained for 165,450 infants with 188,253 admissions.

A complete record of neonatal care was obtained for 94.7% of infants, 76.0% of infants in the data set were in LSOAs where the closest unit of each type (NICU, LNU and SCU) was in the data set and 72.5% of infants had a complete record and were in LSOAs where the closest unit of each type (NICU, LNU and SCU) was in the data set.

In order to minimise the risk of bias due to missing data from units that did not give permission to use data, demand and LOS analysis was based on the 72.5% of infants ($n = 119,967$) who had a complete record and were in LSOAs with all closest unit types represented in the data.

Adjustment of length of stay

The NNRD data are summarised for each day of care. Total care days add up to more than the actual LOS as any part day is counted as a full day in these raw data. We therefore adjusted the days in each level of care in proportion to the total LOS calculated from admission and discharge times (*Figure 1*). The level used in this study was the BAPM 2011 level of care.[2]

Geographic data coverage and predicting neonatal demand

The birth model was based on HES data; therefore, our models do not include the small proportion (≈2%) of births that take place at home.[43] Only 2% of births in England take place in freestanding midwife-led units,[24] and, although these will be in the HES data set, we do not seek to model freestanding midwife-led units as they are currently such a small part of the neonatal care scene.

Permission was given to use data from all NICUs and 85% and 90% of LNUs and SCUs, respectively. Overall, 90% of units gave permission to use their neonatal care data. Because data are missing from LNUs and SCUs, there is the possibility of bias in the statistics used that underlie the model. In order to minimise any bias, statistical summaries were based on only those infants with a complete data record and who live in an area where all closest levels of care locations are present within the data set. Infants whose closest Level 2 or 3 care locations are not in the data set are not used in the analysis (this does not apply to Level 1 NICU care as all NICUs are present in the data set).

As neonatal demand was not available for all hospitals and LSOAs, a regression model was used to predict demand per LSOA. This model was based on LSOAs that had all closest unit types present in the neonatal data set. These represent 75.4% of all LSOAs in England. A demand regression model for neonatal admissions based on births is also less likely to overfit to very specific geographic neonatal demand patterns that may be present simply because of the lower numbers of admissions per geographic area.

A regression model was built based on births and IMD scores. IMD score was chosen as an independent variable because a link between social deprivation and incidence of preterm births has been demonstrated.[44]

The regression model predicted total neonatal admissions for each LSOA. The admissions of VLBW infants and all intensive care, high-dependency care and special care admissions were based on a fixed proportion of total predicted neonatal admissions, based on the number of VLBW infants and use-of-care levels (see *Chapter 5*, *Use-of-care levels and length of stay by gestational age*).

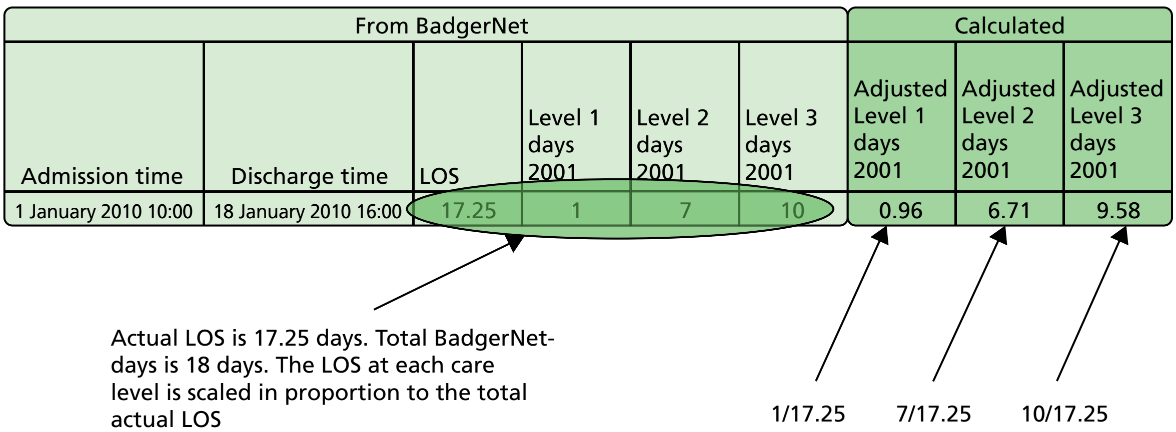


FIGURE 1 Calculation of LOSs in different care levels.

Table 1 shows the coefficients obtained from the regression analysis. Neonatal demand is positively correlated with births and IMD score.

Figure 2 shows the results of the regression analysis when predicting demand at the upper-layer super output area (there are 325 upper-layer super output areas in England). The prediction of demand for VLBW infant admissions, intensive care admissions and high-dependency care admissions all produced correlations with the R^2 of > 90%, showing good regional consistency in predicting these levels of demand. Prediction of special care admissions had an R^2 of 78%. The slightly lower predictive accuracy of special care admissions may be attributable to more regional variation in the clinical judgement on whether or not an infant needs admitting to neonatal care (with much less variation in the perceived need for neonatal care for higher levels of care).

TABLE 1 Regression coefficients for predicting neonatal demand

Term	Coefficient	SE coefficient	*t*-value	*p*-value
1 year births	0.115751	0.000894	129.46	0.00000
IMD score	0.008895	0.000716	12.43	0.00000

SE, standard error.
Notes
Regression was based on births, IMD score and neonatal admissions at LSOA level.
The adjusted R^2 for all admissions was 74.0%.

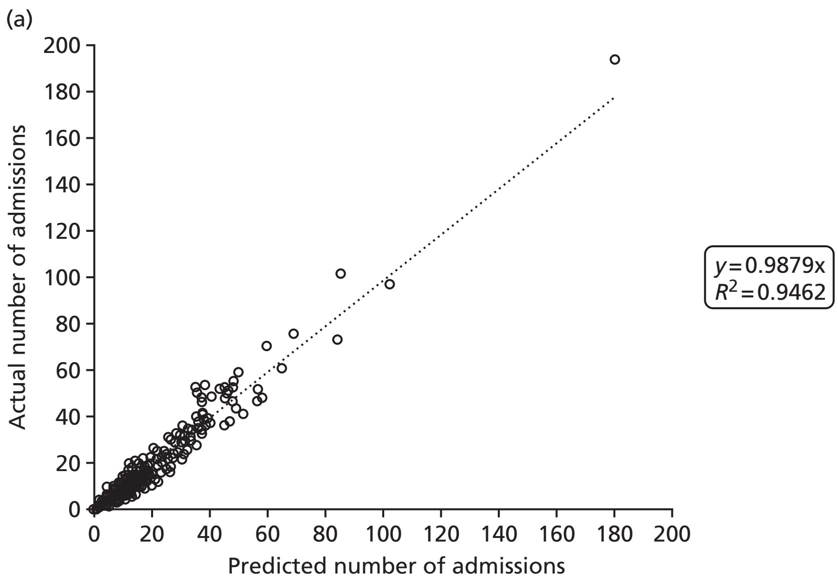


FIGURE 2 Correlation between actual and predicted neonatal admissions at upper-layer super output area. (a) VLBW admissions; (b) infants with ≥ 24 hours of intensive care; (c) infants with ≥ 24 hours of high-dependency care; and (d) infants with ≥ 24 hours of special care. (*continued*)

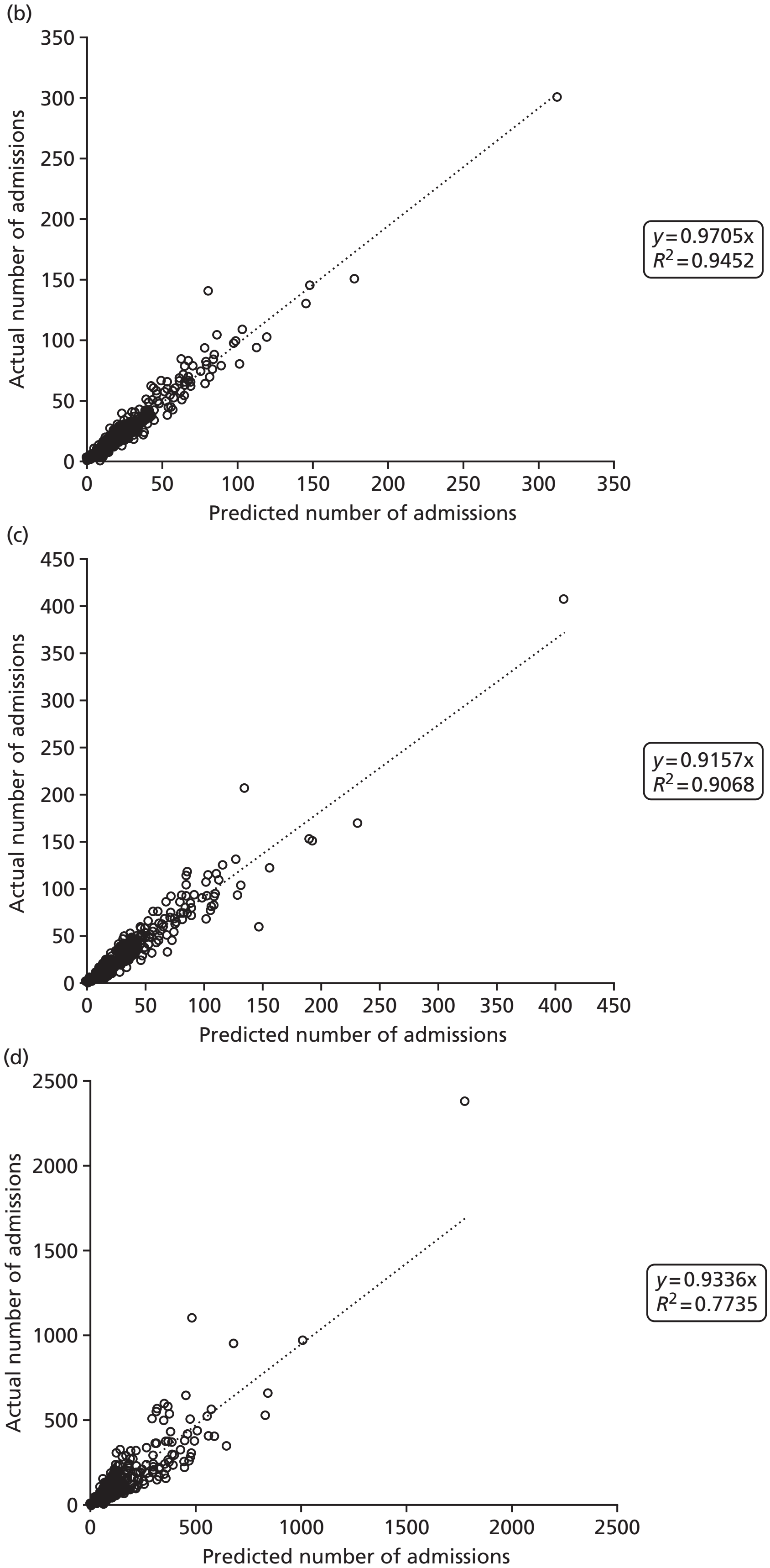


FIGURE 2 Correlation between actual and predicted neonatal admissions at upper-layer super output area. (a) VLBW admissions; (b) infants with ≥ 24 hours of intensive care; (c) infants with ≥ 24 hours of high-dependency care; and (d) infants with ≥ 24 hours of special care.

Chapter 5 Descriptive analysis of data

This chapter provides a general descriptive analysis of factors pertinent to neonatal workload, LOS, mortality, travel times and type of unit where care is received.

Data used for descriptive analysis

As described in *Chapter 4*, *Neonatal data*, only those infants with a full neonatal record and whose mothers live in areas where all closest levels of care are present in the data set are used. Analysis is based on 119,967 infants. Definitions of intensive care, high-dependency care and special care are in accordance with the BAPM 2011 guidelines.[2]

Gestational age is given as completed weeks of gestation; for example, a gestational age of 28 weeks includes those infants with a gestational age of 28^{+0} to 28^{+6} (inclusive).

Calculation of nurse workload

Nurse workload is calculated in accordance with BAPM standards of care,[2] in which one nurse is recommended to care for one infant in intensive care (Level 1), two infants in high-dependency care (Level 2) or four infants in special care (Level 3). No guidelines exist for nurse requirements for transitional care (Level 4). In this study, we have assumed the same mathematical progression that is seen between other levels of care and assume that one nurse could care for eight infants in transitional care. Calculated nurse workload is for those nurses involved in direct infant care only (nurses in charge of shift, managing nurses and nurses dedicated to infant transfer are not included). Nurse workload was calculated as:

$$\text{Nurse workload} = \text{Infant}_{L1} + \frac{\text{Infant}_{L2}}{2} + \frac{\text{Infant}_{L3}}{4} + \frac{\text{Infant}_{L4}}{8}, \quad (1)$$

where:

- Nurse workload = nurse required (BAPM 2011 guidelines[2])
- Infant_{L1} = infant in Level 1 (intensive) care
- Infant_{L2} = infant in Level 2 (high-dependency) care
- Infant_{L3} = infant in Level 3 (special) care
- Infant_{L4} = infant in Level 4 (transitional) care.

Admissions, bed-days and nurse workload by gestational age at birth

Table 2 shows admissions, bed-days and nurse workload by gestational age. Fifty per cent of admissions are up to 37 weeks' gestational age at birth, but 50% of beds are occupied by infants with a gestational age at birth of ≤ 32 weeks, and 50% of nurse workload is occupied with infants with a gestational age at birth of ≤ 31 weeks.

TABLE 2 Admissions, bed-days and nurse workload by gestational age at birth

Gestational age (weeks)	%			Cumulative %		
	Admissions	Bed-days	Nurse workload	Admissions	Bed-days	Nurse workload
22	0.01	0.03	0.05	0.01	0.03	0.05
23	0.25	1.30	2.36	0.26	1.32	2.41
24	0.44	3.34	5.46	0.70	4.66	7.87
25	0.51	3.96	6.01	1.20	8.62	13.88
26	0.64	4.65	6.45	1.84	13.27	20.33
27	0.77	5.16	6.75	2.61	18.43	27.08
28	1.08	6.41	7.81	3.69	24.84	34.89
29	1.28	6.21	6.82	4.97	31.05	41.71
30	1.69	6.68	6.49	6.66	37.73	48.20
31	2.27	6.98	6.36	8.93	44.70	54.56
32	3.19	7.40	6.20	12.12	52.11	60.77
33	4.46	7.43	6.00	16.58	59.53	66.77
34	7.30	8.23	6.26	23.88	67.76	73.03
35	7.25	5.54	4.41	31.13	73.30	77.43
36	9.25	4.82	3.91	40.38	78.12	81.34
37	10.37	4.47	3.80	50.74	82.59	85.15
38	10.50	4.01	3.49	61.25	86.59	88.64
39	12.34	4.27	3.65	73.58	90.86	92.29
40	13.86	4.74	4.00	87.44	95.60	96.29
41	10.64	3.73	3.17	98.07	99.33	99.46
42	1.90	0.66	0.54	99.98	99.99	100.00

Mortality by gestational age

Mortality by gestational age at birth is shown in *Table 3*. Mortality reduced from 86% at a gestational age of 22 weeks at birth to 7% at a gestational age of 28 weeks at birth; however, because of larger numbers of births at higher gestational ages, 50% of all mortality still occurred in infants with a gestational age of ≥ 28 weeks at birth (these deaths include all causes of neonatal death).

Use-of-care levels and length of stay by gestational age

Table 4 shows use-of-care levels and LOS by gestational age at birth. Of those born at a gestational age of < 32 weeks, more than half will require a period in intensive care.

On average, for all neonatal admissions, one infant uses 1.4 days of Level 1 care, 2.0 days of Level 2 care, 7.3 days of Level 3 care and 0.3 days of Level 4 care; this totals 11.0 days of care. The total nurse workload days per infant averages 4.3 nurse-days.

Use of beds and nurse resources is shown in *Figure 3*.

TABLE 3 Mortality and cumulative mortality by gestational age at birth

Gestational age at birth (weeks)	Mortality	
	%	Cumulative %
22	85.71	0.77
23	63.39	12.76
24	40.04	26.28
25	22.28	34.94
26	15.47	42.50
27	8.37	47.44
28	6.55	52.88
29	3.52	56.35
30	2.16	59.17
31	1.87	62.44
32	1.20	65.38
33	0.77	68.01
34	0.62	71.47
35	0.52	74.36
36	0.55	78.27
37	0.51	82.37
38	0.57	86.99
39	0.52	91.92
40	0.40	96.15
41	0.44	99.74
42	0.18	100.00

TABLE 4 Use-of-care levels and LOS by gestational age at birth

Gestational age at birth (weeks)	Use-of-care levels (% of all infants)				LOS when level used (days)				LOS, using a value of 0 when level unused (days)				
	L1	L2	L3	L4	L1	L2	L3	L4	L1	L2	L3	L4	Σ
22	100.0	14.3	21.4	7.1	12.4	75.2	4.3	0.4	12.4	10.7	0.9	0.0	24.1
23	100.0	41.7	38.0	2.4	28.3	50.0	23.3	3.1	28.3	20.8	8.9	0.1	58.1
24	100.0	65.7	60.2	7.0	30.9	54.5	27.9	3.7	30.9	35.8	16.8	0.3	83.7
25	99.7	81.2	79.9	11.2	27.0	44.4	28.8	2.6	27.0	36.1	23.0	0.3	86.3
26	99.3	87.2	84.7	14.8	20.2	37.4	32.4	2.7	20.1	32.6	27.5	0.4	80.5
27	99.2	92.7	90.8	19.5	16.8	28.5	33.5	2.7	16.7	26.4	30.4	0.5	74.0
28	96.9	91.3	92.7	21.3	13.4	21.1	35.0	2.8	13.0	19.3	32.4	0.6	65.3
29	94.4	92.1	95.8	25.7	9.3	12.5	33.9	2.6	8.8	11.5	32.5	0.7	53.4
30	76.4	86.5	98.0	25.5	6.3	9.0	30.8	2.5	4.8	7.8	30.2	0.6	43.4

continued

TABLE 4 Use-of-care levels and LOS by gestational age at birth (*continued*)

Gestational age at birth (weeks)	Use-of-care levels (% of all infants)				LOS when level used (days)				LOS, using a value of 0 when level unused (days)				
	L1	L2	L3	L4	L1	L2	L3	L4	L1	L2	L3	L4	Σ
31	59.3	80.4	98.3	25.6	5.2	6.4	25.4	2.5	3.1	5.1	25.0	0.6	33.8
32	36.6	66.7	98.7	27.8	4.3	4.9	20.3	2.4	1.6	3.3	20.1	0.7	25.6
33	23.9	50.9	99.0	25.2	3.9	4.1	14.9	2.1	0.9	2.1	14.7	0.5	18.3
34	14.5	34.4	98.4	22.5	3.0	3.4	10.5	2.1	0.4	1.2	10.4	0.5	12.4
35	11.7	24.9	97.8	20.8	3.7	3.5	6.8	2.0	0.4	0.9	6.7	0.4	8.4
36	10.1	17.9	96.7	17.6	3.3	3.5	4.6	1.8	0.3	0.6	4.4	0.3	5.7
37	11.0	16.8	96.0	15.1	3.3	3.1	3.8	1.5	0.4	0.5	3.6	0.2	4.7
38	10.5	13.6	95.6	13.0	3.6	3.2	3.4	1.3	0.4	0.4	3.2	0.2	4.2
39	9.4	12.6	95.9	11.7	3.5	2.8	3.1	1.3	0.3	0.3	3.0	0.1	3.8
40	10.1	12.0	96.5	10.5	3.1	2.3	3.1	1.3	0.3	0.3	3.0	0.1	3.8
41	10.7	12.9	96.9	10.2	3.2	2.1	3.2	1.2	0.3	0.3	3.1	0.1	3.9
42	9.3	11.7	97.0	9.3	2.9	2.0	3.3	1.3	0.3	0.2	3.2	0.1	3.9

L, level.

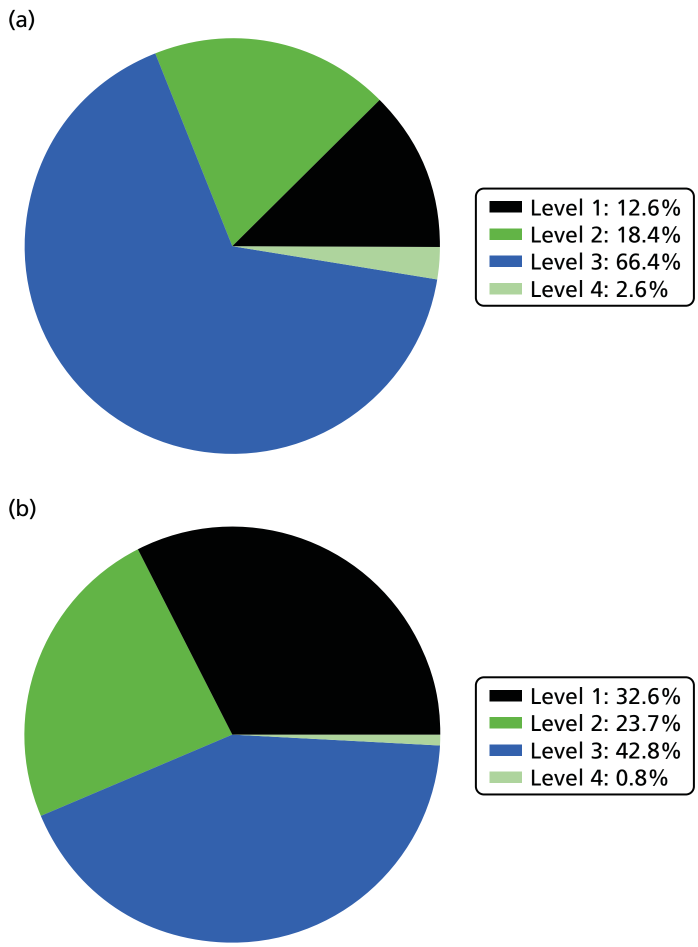


FIGURE 3 Use of beds and nurse resources by BAPM care level. (a) Bed use; and (b) nurse workload.

Overall, 1.29% of infants cared for in neonatal units required specialist surgical care carried out in a NICU (when infants with incomplete BadgerNet records, which includes infants who travel to surgical units independent of NICUs, were included in the analysis, this figure rose to 2.45%). This was dependent on gestational age at birth: at 23–25 weeks, 10–13% of infants required specialist surgical care (in a NICU/surgical centre); this fell to 1–2% of infants at 31–33 weeks and < 1% of infants at ≥ 34 weeks.

Gestational age at discharge

Figure 4 shows gestational age at discharge by gestational age at birth. The lowest gestational age at discharge is for infants who are born between 30 and 35 gestational weeks (discharged, on average, at a gestational age of ≈36 weeks). A low gestational age at birth (≤ 25 weeks) is associated with an average gestational age at discharge of > 40 weeks.

Travel time and distance to place of care

The mean distance from the mother's home to the place of care is shown in *Figure 5*.

As not all neonatal units provide all levels of care, travel times can depend on the level of care required. A spell refers to the time spent by an infant at one particular neonatal unit.

Spells involving Level 1 (intensive) care had a median travel time from home of 21 minutes, and a median distance from home of 8 miles. Ten per cent of spells were > 61 minutes and > 42 miles from home.

Spells involving Level 2 (high-dependency) care had a median travel time from home of 16 minutes, and a median distance from home of 6 miles. Ten per cent of spells were > 46 minutes and > 28 miles from home.

Spells involving Level 3 (special) care had a median travel time from home of 13 minutes, and a median distance from home of 5 miles. Ten per cent of spells were > 31 minutes and > 18 miles from home.

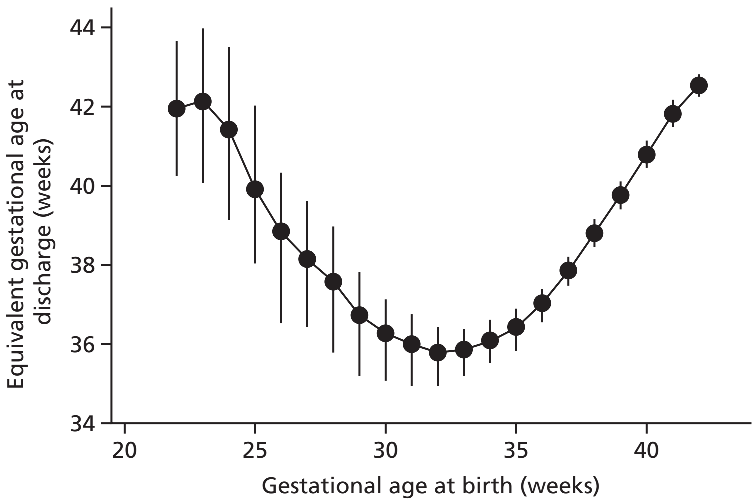
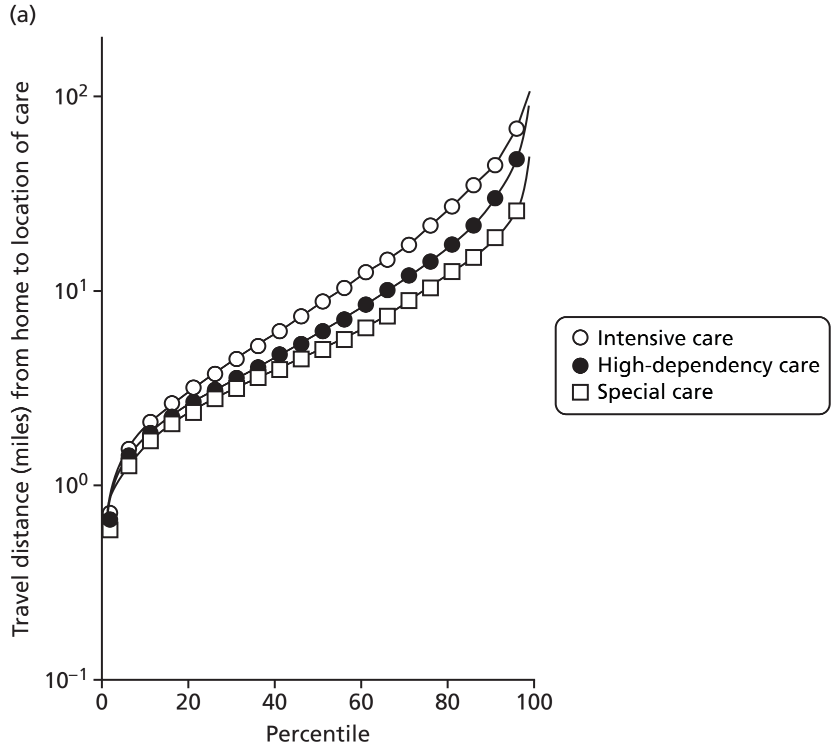


FIGURE 4 Gestational age at discharge by gestational age at birth. Points represent means and bars represent standard deviations.



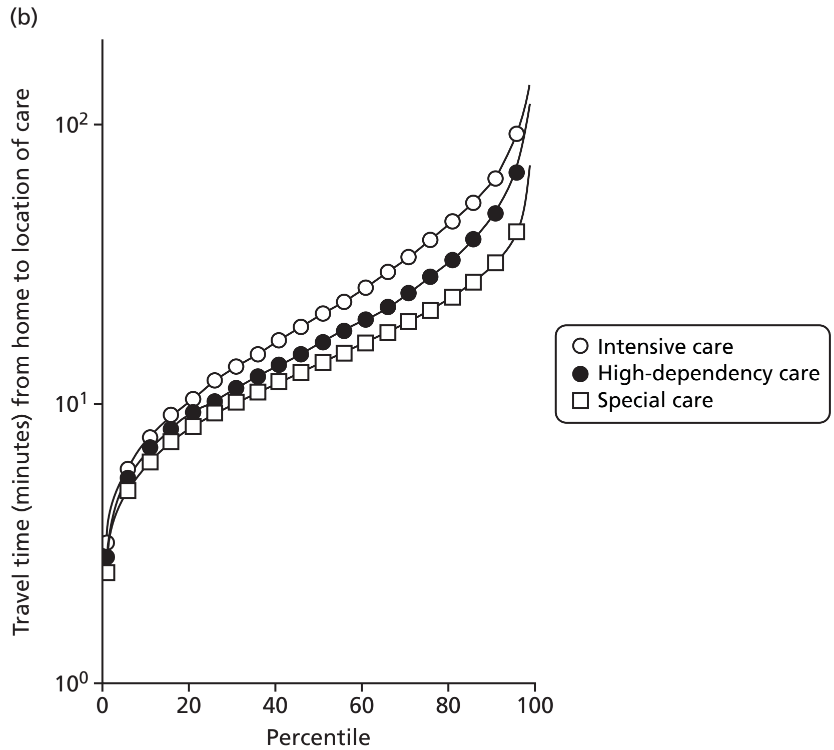


FIGURE 5 Mean travel (a) distance (miles); and (b) time (minutes) per spell by level of care.

The data were analysed for infants who received care for > 1 day at a unit that was > 15 minutes further away than their closest appropriate unit:

- 12.9% of spells involving a Level 1 stay of > 1 day were > 15 minutes further away than their closest appropriate unit. On average, the excess travel time was 40 minutes and the average excess duration of stay was 10.6 days.
- 14.1% of spells involving a Level 2 stay of > 1 day were > 15 minutes further away than their closest appropriate unit. Of those not requiring Level 1 care (and so would not be waiting for transfer back from a higher-level unit), 7.9% were cared for at a unit > 15 minutes further away than their closest appropriate unit, with an average excess travel time of 41 minutes and an average excess duration of stay of 5.3 days.
- 6.7% of spells involving a Level 3 stay of > 1 day were > 15 minutes further away than their closest appropriate unit. Of those not requiring Level 1 or 2 care (and so would not be waiting for transfer back to a SCU from a higher-level unit), 4.7% were cared for at a unit > 15 minutes further away than their closest appropriate unit, with an average excess travel time of 38 minutes and an average excess duration of stay of 6.0 days.

Hospital birth by neonatal unit type

An analysis was performed on place of birth and the level of neonatal unit available at the place of birth.

For all infants who have ≥ 1 day of BAPM Level 1 (intensive) care:

- 53.0% are born in a hospital with a NICU
- 18.1% are born in a hospital without a NICU and are transferred to a NICU
- 28.9% are born in a hospital without a NICU and are not transferred to a NICU.

For all infants who have ≥ 7 days of BAPM Level 1 (intensive) care:

- 60.4% are born in a hospital with a NICU
- 20.8% are born in a hospital without a NICU and are transferred to a NICU
- 18.9% are born in a hospital without a NICU and are not transferred to a NICU.

For all infants with a gestational age of < 33 weeks at birth and who have ≥ 7 days of BAPM Level 1 (intensive) care:

- 61.2% are born in a hospital with a NICU
- 18.9% are born in a hospital without a NICU and are transferred to a NICU
- 19.8% are born in a hospital without a NICU and are not transferred to a NICU.

For all infants with a gestational age of < 28 weeks at birth and who have ≥ 7 days of BAPM Level 1 (intensive) care:

- 72.0% are born in a hospital with a NICU
- 20.9% are born in a hospital without a NICU and are transferred to a NICU
- 7.1% are born in a hospital without a NICU and are not transferred to a NICU.

Chapter 6 Location analysis

In acute health-care settings, there can often be a tension between providing local services close to the patients and developing centres of excellence with extensive experience gained through high volumes of specialist work. This chapter examines the compromise between patient travel time and unit size, and analyses the optimal locations for birth centres and neonatal care units.

Model

We consider the space of possible locations given by the existing H neonatal units of England. Given a number of $h \in [\![1; H]\!]$ available units, we wanted to find the optimal combination of unit locations to optimise a set of criteria. Such combinations are described by vectors, $u = \{u_1, u_2, \ldots, u_h\}$, where $\forall i \in [\![1; h]\!]$ $u_i \in [\![1; H]\!]$ without repetition. The set of possible combinations is called the feasible set of the decision space.

Assumptions

We assume that all units have an unlimited capacity. However, the maximum number of admissions to any single unit is kept as low as possible by criterion 6 stated in *Decision criteria*.

We assume that neonatal care can only take place in the existing neonatal units. Considering that the existing network is already quite dense and the tendency is towards a reduction of the number of units, then this assumption is reasonable.

Decision criteria

The aim of this study is to optimise the location of neonatal units in order to improve the outcome of the infants, the experience of the parents and to align the neonatal network with the current demand. These objectives translate into the following criteria:

1. Minimise the average distance from a mother's place of residence to an available unit.
2. Minimise the maximum distance for any mother to an available unit.
3. Maximise the proportion of mothers living within 30 minutes of the nearest available unit (other travel time limits, such as 45 and 60 minutes, are also evaluated, but the 30-minute range is used for optimisation as this range is more discriminating between options).
4. Maximise the proportion of births taking place in units with more than a given number, A_{min}, of admissions per year.
5. Maximise the minimum number of admissions (for any single unit) below a given number of admissions per year.
6. Minimise the maximum number of admissions (for any single unit) over a given number of admissions per year.
7. Maximise the proportion of mothers within 30 minutes of the nearest available unit and going to a unit with more than a given number, A_{min}, of admissions per year.

The first three coverage criteria, very common in facility location problems, aim to align facility locations with the population distribution. They rely on the assumption that women will go the nearest care facility.[45] The other four criteria are based on the number of admissions to each unit, once every woman has been assigned to an available location. In particular, criterion 4 is specific to this study and favours the creation of big units in order to improve clinical outcomes.[22] Criteria 3 and 4 can conflict in areas with a sparse population. Finally, criterion 6 limits the maximum size of care units. Possible criteria values define the objective space.

Dealing with multiple criteria: Pareto dominance

Our study tackles a multiobjective optimisation (MOO) problem. When solving an optimisation problem based on one objective, the optimal solution is given by the configuration with the best (highest or lowest) objective value. In the case of MOO, comparing several solutions requires reference to the notion of dominance: a vector a of the objective space dominates another vector b if all criteria of a are better or equal to the criteria of b and $a \neq b$.[46] Then, there is no single best solution but a set of non-dominated solutions, called the Pareto front.

Complexity of the decision space

If the problem is to find the best combination of h units in a list of H possible locations, then one common method is to compute all objectives for all possible combinations (brute force). The number of h-combinations (combinations of size h) is the binomial coefficient:

$$\binom{H}{h} = \frac{H!}{h!(H-h)!}, \tag{2}$$

if $h \leq H$, or equals zero if $h > H$. With $H = 161$ possible locations, there are, for instance, 12,880 combinations of two locations and the maximum number of configurations is 1.82×10^{47}, reached with half of locations (80) available. With such a large number of combinations, the brute force method cannot be used and the optimisation problem must be solved by heuristic methods. In practice, the size of the decision space means that it is impossible to give an exact solution to this optimisation problem; only an approximate solution can be given.

Optimisation methods in the literature

This study tackles a facility location problem, with several deterministic objective functions. The decision space contains a finite number of potential locations, so the optimisation problem is combinatorial.

Our aim was to find the best h-combinations for all $h \in [\![1; H]\!]$ that maximise the objective functions. In practice, it means finding the Pareto front of non-dominated solutions. As the optimisation problem is NP (non-deterministic polynomial time) hard, there is no known method to find the exact solution or true Pareto front of this problem in a reasonable time;[47] however, the literature offers a lot of heuristic methods that can provide approximate solutions.

According to Berman *et al.*,[48] large facility location problems can be solved by descent algorithms, simulated annealing and genetic algorithms. Genetic algorithms were shown to produce the best results but are more computationally expensive.[48]

Greedy algorithm

The greedy algorithm is a deterministic technique that leads to one local optimum. The greedy algorithm cannot identify a Pareto front, but seeks to find the best solutions based on a composite score (the 'fitness value') of all objectives. The solution identified is highly dependent on how the different objectives are weighted. The algorithm starts with the location with the highest fitness value. At each iteration, the algorithm selects the location that brings the most improvement and adds it to the combination. Although this technique is intuitive and fast, it provides only one solution and it strongly relies on weights and fitness.

Steepest-ascent hill climbing

The hill-climbing method is an efficient method to find local optima of the fitness function. From a given individual, candidates are generated by mutating only one gene of the original. Then, the fitness value for each candidate is computed. The individual is then replaced by the candidate that brings the highest improvement (steepest ascent). If no improvement is possible, the local optimum is recorded and the exploration is restarted from another random point.

Simulated annealing

Simulated annealing is a stochastic local search method.[47] Here, annealing refers here to the cooling of materials with a controlled temperature. The probability of accepting combinations with a worse fitness decreases with the temperature, so that the search converges to an optimum.

Genetic algorithms

Genetic algorithms manage a population of individuals encoded as vectors through a given number of generations. At each generation, 'good' parents are selected from the population depending on their fitness. Parents are then combined, using a crossover operator, to create children that are finally mutated. Genetic algorithms differ in the parent selection process, in the crossover and mutation processes, and in the way that the population is archived.

Selection

The selection operator chooses a part of the population to become parents. The better individuals in terms of objective values are more likely to become parents.

The selection probability can be proportionate to fitness by roulette-wheel sampling[49] or stochastic universal sampling.[50] The sigma scaling method normalises the fitness by its variance in the population, so that the individuals with the highest fitness always have a higher probability than others to produce children. However, these approaches focus on exploitation of an existing population rather than exploration of the decision space, and they can lead to premature convergence.

Other selection methods rely on ranking rather than fitness value. With ranking selection, individuals are ranked depending on their fitness, and their probability to become parents is a function of their rank.[51] Similarly, the tournament selection picks random pairs of individuals and determines which has the highest fitness value. The individual with a higher fitness will be selected with a given probability (e.g. if probability is set at 0.7 then the individual with higher fitness will be selected 70% of the time, and the individual with lower fitness will be selected 30% of the time).[52] Such methods allow the algorithm to keep some individuals with low fitness values (with the advantage of keeping a broader gene pool).

Finally, the Boltzmann selection[53] controls the selection rate via a temperature. At the beginning, all individuals have a similar probability to be selected. As the temperature decreases, the selection focuses on high-fitness individuals.

Crossover

The crossover is the process that exchanges genes from parents to create new children. The simplest option is the single-point crossover, which selects one locus and exchanges blocks before and after that locus; for example, we may code whether an individual hospital is open with 0 (closed) or 1 (open). The open/closed status of eight hospitals is given as a vector, such as 00000000 (all closed), 11111111 (all open) or 10000001 (hospitals 1 and 8 are open, and the rest are closed). With crossover, two different configurations (parents) are mixed by exchanging at a random locus within the vector; for instance, mixing vectors 00001111 and 01010101 after the fifth locus gives the children 00001101 and 01010111. The choice of the single-point location can be made by a uniform distribution. In the case of binary vectors, the single-point crossover is less likely to exchange the endpoints of vectors.[54] To reduce this effect, the crossover can rely on two or more exchange points. In the case of integer vectors, an additional repair step is necessary to remove any potential repetition.

Mutation

Mutation changes the gene value of each locus, with a very small probability for each individual generation. According to Holland,[55] the mutation process avoids the loss of diversity in the population.

Archive

Genetic algorithms also vary by the way solutions are archived and if the population size is variable. The simple option is to keep only children; however, it assumes that children are better than parents that are lost. Several methods build an archive that is a union of parents and children. If the population size is variable, an option is to keep the Pareto front of this archive; however, the size of this Pareto front can increase dramatically, in particular with many objective functions. Then, individuals from the archive are ranked, based on their Pareto dominance and another metric. Non-Sorting Genetic Algorithm II (NSGA-II)[56] and SPEA (Strength Pareto Evolutionary Algorithm) 2[57] both rank individuals by combining dominance and spread metric in order to maximise population diversity.

The Non-Sorting Genetic Algorithm II method

In NSGA-II,[56] the archive and the new population are merged and all individuals are ranked in accordance with a two-step mechanism. In the first step, the merged population is split into layers of non-dominated fronts, the first layer being the Pareto front (the second layer being the next Pareto front after removal of the first layer). In the second step, the spread of the population is measured by the crowding distance, which gives the distance from an individual to its nearest neighbour. To keep the size of the population constant, a given number of individuals is selected from the merged population, preferably from the upper layers and with the largest crowding distance.

The NSGA-II has the opportunity to keep not only optimal solutions, but also near-optimal solutions, in lower layers; however, to do so, the population must be large enough. Another advantage is to provide a diverse population in terms of score values, thanks to the crowding-distance ranking.

The NSGA-II was chosen for this study after a comparison with SPEA 2,[57] MOEAD (multiobjective evolutionary algorithm based on decomposition),[46] and HypE (hypervolume estimation algorithm),[58] which showed that NSGA-II provided similar objective performances with a more diverse population.

Convergence metrics

The number of generations was determined with a steady-state detection-based termination criterion (a chi-squared test based on the generational distance of objective values, or on the Hamming distance of integer vectors) adapted from Wong *et al.*[59]

Maternity unit location analysis

This section focuses on the centralisation of maternity units in England. The relationship between the number and location of obstetric units and the access of care quantified by the model criteria (see *Decision criteria*) is analysed to measure the impact of a potential reorganisation of care.

Data

Data on the number of births in existing maternity units were provided by the HES database (see *Chapter 4*, *Birth data*). Birth numbers per year were averaged using data from 2013 to 2015.

Travel times for patients between the LSOAs of England (country divisions with similar population sizes as defined in 2011) and the 161 existing maternity units were estimated (see *Chapter 4*, *Travel time data*). Maternity units were selected from those hospitals that had any level of neonatal care unit.

Estimation

The NSGA-II method[56] and the criterion functions defined in *Decision criteria* were implemented using MATLAB programming software (MathWorks, Natick, MA, USA). In particular, since the Royal College of Obstetricians and Gynaecologists has recommended that units have ≥ 6000 births per year,[22] criteria 4 and 1 were applied with a number of births per year defined by $A_{min} = 6000$.

Criterion functions were normalised between 0 and 1 using values from *Table 5*, with 1 being the ideal value. Fitness was computed as the weighted average of normalised objective values, with equal weights for all objectives.

The first generation was generated randomly using an integer uniform distribution in $[\![1; h]\!]$, so that there was no location repetition inside combination chromosomes and no duplicates in the population.

The population size was set to $P = 200$ and remained constant through generations. This value was chosen as a compromise between the exploration of the decision space and the computing time.

The population was propagated through $G = 200$ generations; this value would provide a steady state as described in *Convergence metrics*.

The MOO process was run independently for all $h \in [\![1; H]\!]$. Note that the same optimisation algorithm can be applied with a binary location encoding $(u_1, \ldots, u_H) \in \{0; 1\}^H$. In that case, the number of open locations is flexible and only one optimisation run provides a Pareto front with various numbers of locations. However, the range of location numbers in the final solutions is not easily controlled with such encoding and depends on score quality. The integer encoding allows a more thorough exploration of the decision space.

Parents were selected from the population using the tournament selection process.[56] To do so, pairs of individuals are randomly drawn from the population and the one with the highest fitness is selected as a parent with a probability of $p_t = 0.75$. The tournament is an elitist selection process as it favours high fitness values but it allows lower fitness values with a probability of $1 - p_t$.

TABLE 5 Criteria for the optimisation of maternity or neonatal unit locations

		Normalisation interval (normalisation range)			
Criterion	Direction	Maternities study	NICUs study	LNUs study	SCUs study
Average travel time from mother to closest unit	Minimum	[15–344] minutes	[14–335] minutes	NA	NA
Maximum travel time from any mother to closest unit	Minimum	[82–570] minutes		NA	NA
Proportion of mothers living within 30 minutes of the closest unit (target 1)	Maximum	[0–0.94]	[0–0.95]	NA	NA
Proportion of admissions in units more than A_{min} per year (target 2)	Maximum	[0.20–1] ($A_{min} = 6000$ births)	[0–1] ($A_{min} = 100$ VLBW)	NA	NA
Minimum number of admissions for any single unit	Maximum	[1100–6000]	[13–100]	[28–15,386]	[127–68,586]
Maximum number of admissions for any single unit	Minimum	[12,000–63,828]	[1000–6806]	[28–15,386]	[127–68,586]
Proportion of mothers and infants meeting targets 1 and 2	Maximum	[0–1]	[0–1]	NA	NA

NA, not applicable.

Parents are then combined using a single-point crossover; the crossover point location is randomly chosen following a uniform distribution.

The alleles of every child were mutated with a probability of $p_m = 0.001$. After crossover and mutations, additional mutations took place to ensure that children did not contain location repetitions.

The outputs of the optimisation process are the final Pareto front layers up to *P* individuals and their corresponding objective values. The minimum number of admissions for each combination is also recorded in order to evaluate the impact of the redistribution of locations.

Results

Access to care

Figure 6a shows the relationship between the average and maximum travel time for mothers, assuming that they go to the closest unit, as a function of the number of available obstetric units. Each dot represents the criterion value of one configuration or set of units discovered in the Pareto front of solutions.

With the current number of 161 obstetric units, the average travel time is 15 minutes and the maximum travel time is 82 minutes. As expected, the travel time increases when the number of units decreases; for instance, reducing the number of units by half to 80 increases the average travel time to 21 minutes (+6 minutes) and the maximum travel time to 99 minutes (+17 minutes). Note that removing one unit only affects the patient access in some LSOAs and the average travel time is computed for all LSOAs. As a consequence, the effect on the average travel time is limited.

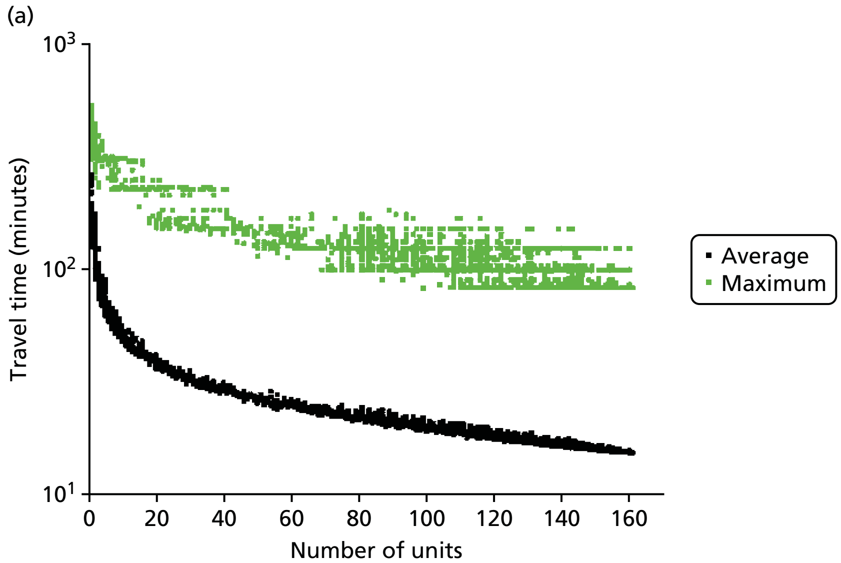
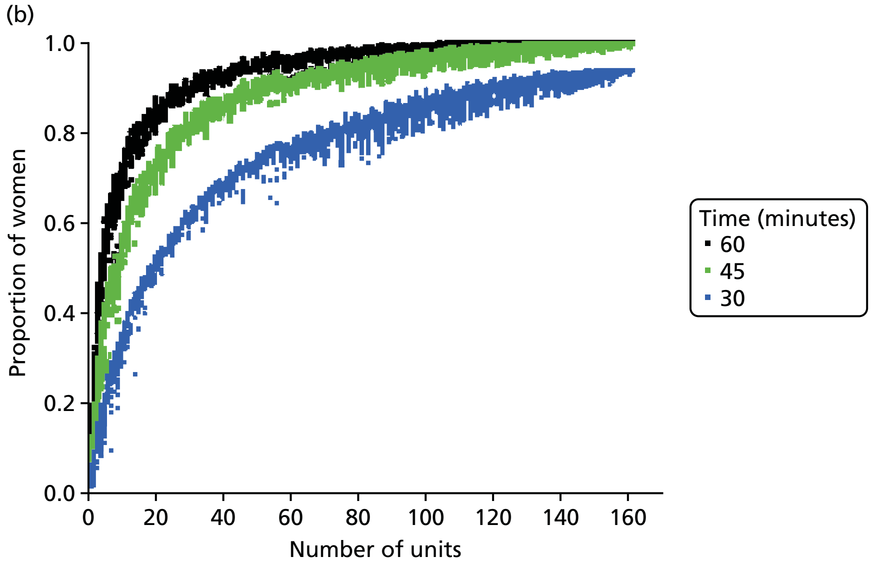


FIGURE 6 Influence of the number of maternity units on the access to care. (a) Travel time (minutes) (potential configuration: minimum and maximum travel times); (b) proportion of women in target time (potential configuration: proportion of women within 30, 45 or 60 minutes of a maternity unit); (c) number of admissions (potential configuration: minimum and maximum admissions to any single unit); (d) births in units with ≥ 6000 births per year (%) (potential configuration: proportion of births in a unit with ≥ 6000 admissions per year); and (e) proportion of births in target (potential configuration: the proportion of births within 30 minutes, proportion of births in a unit with ≥ 6000 admissions per year, and proportion of births in a unit with 6000 births per year and within 30 minutes). Quantified by the distance to units and the proportion of women in target time, on the number of admissions and on the proportion of births occuring in large units (≥ 6000 births per year). (*continued*)



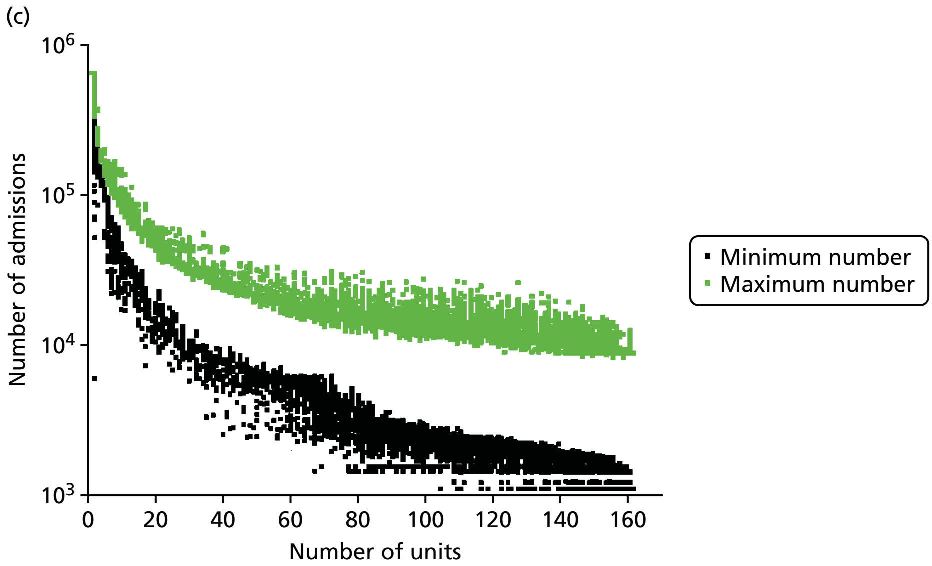
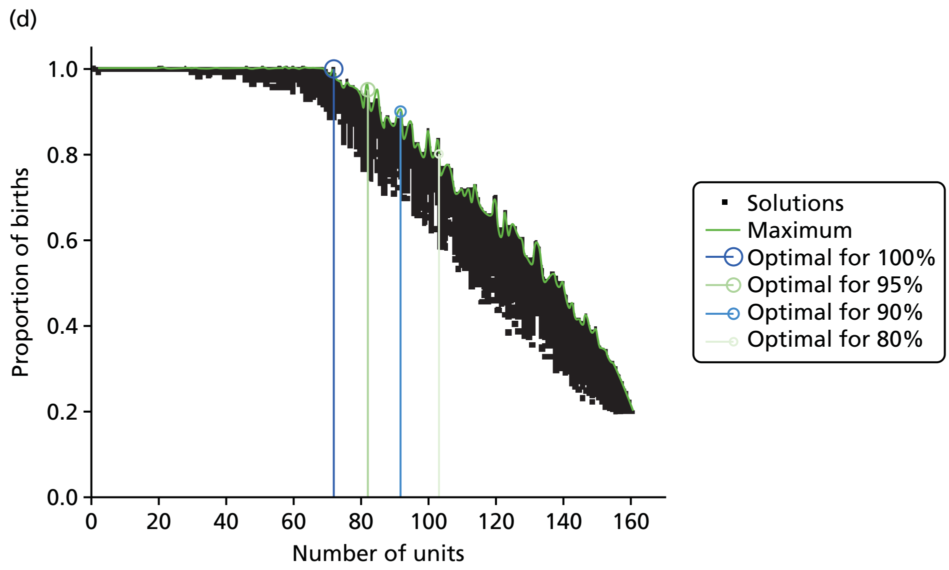


FIGURE 6 Influence of the number of maternity units on the access to care. (a) Travel time (minutes) (potential configuration: minimum and maximum travel times); (b) proportion of women in target time (potential configuration: proportion of women within 30, 45 or 60 minutes of a maternity unit); (c) number of admissions (potential configuration: minimum and maximum admissions to any single unit); (d) births in units with ≥ 6000 births per year (%) (potential configuration: proportion of births in a unit with ≥ 6000 admissions per year); and (e) proportion of births in target (potential configuration: the proportion of births within 30 minutes, proportion of births in a unit with ≥ 6000 admissions per year, and proportion of births in a unit with 6000 births per year and within 30 minutes). Quantified by the distance to units and the proportion of women in target time, on the number of admissions and on the proportion of births occuring in large units (≥ 6000 births per year). (*continued*)



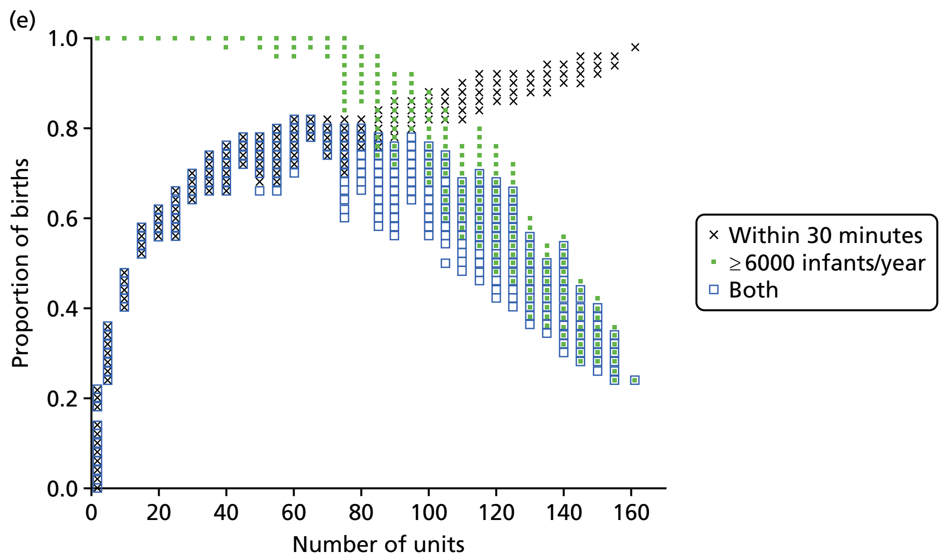


FIGURE 6 Influence of the number of maternity units on the access to care. (a) Travel time (minutes) (potential configuration: minimum and maximum travel times); (b) proportion of women in target time (potential configuration: proportion of women within 30, 45 or 60 minutes of a maternity unit); (c) number of admissions (potential configuration: minimum and maximum admissions to any single unit); (d) births in units with ≥ 6000 births per year (%) (potential configuration: proportion of births in a unit with ≥ 6000 admissions per year); and (e) proportion of births in target (potential configuration: the proportion of births within 30 minutes, proportion of births in a unit with ≥ 6000 admissions per year, and proportion of births in a unit with 6000 births per year and within 30 minutes). Quantified by the distance to units and the proportion of women in target time, on the number of admissions and on the proportion of births occuring in large units (≥ 6000 births per year).

In *Figure 6b*, the proportion of women living within 30/45/60 minutes of the closest obstetric unit increases with the number of units. With 161 units, 93% of women live within 30 minutes and 100% live within 45 minutes of the closest unit. With only half of the units (80), the proportion of women (1) within 30 minutes of the closest unit is reduced to 84% (–9%), (2) within 45 minutes is reduced to 90% (–10%) and (3) within 60 minutes is reduced to 95% (–5%). Similar to the average travel times, the effect of changing the number of units on the proportion of women within a target travel time is rather smooth because it only affects a part of the population.

Size of units

The number of admissions per unit decreases when the number of units increases, as shown in *Figure 6c*. With 161 units, the number of admissions ranges from 1100 to 8743. With only half of the units (80), the highest estimated minimum of admissions is 4562 and the lowest estimated maximum number is 11,960; however, both values may not be achievable in the same configuration. To achieve a minimum number of 6000 births per year, the highest number of units is estimated to be 72 units.

Figure 6d represents the proportion of births occurring in units with ≥ 6000 births per year as a function of the number of obstetric units. With the current configuration of 161 units, only 20% of births occur in units with ≥ 6000 births per year (or 'large units'), in accordance with our model. Reducing the number of units by 21, to 140 units, could lead to an increase of 30%, to reach 50%. Thus, the relationship between the number of units and the proportion of births in large units is particularly strong and a small change can lead to a big impact. Furthermore, to reach a proportion of 80%/90%/95%/100%, the number of units would need to be reduced to approximately 103/92/82/72 units, respectively.

Compromise

There is trade-off in the number of units to achieve both a high proportion of mothers within a target travel time and a high proportion of births in units with ≥ 6000 births per year. *Figure 6e* shows the proportion of patients within 30 minutes of their nearest unit and attending large units, as a function of the number of units. By reducing the number of units from 161 to approximately 65 (±5), the proportion would be increased from 24% to 82%, the maximum achievable based on our results. Note that it is not possible to achieve 100% of patients for both targets because there are not enough patients.

Regional population projections

The 10-year projection for changes in women of child-bearing age (considered to be those aged 15–39 years) ranged from a reduction of about 1% in the North West to an increase of about 4% in London (*Table 6*).

These projections were made before the UK voted to leave the European Union (expected to formally take place in March 2019). There may, therefore, be significant uncertainty about these projections as the age group of interest coincides with the mobile working-age population. Owing to current uncertainty, we have not sought to build projections into our modelling; rather, when considering the output of the modelling, it should be remembered that admission numbers may change to be between 1% lower and 4% higher over the course of 10 years.

Neonatal intensive care location analysis

Neonatal care units are organised in three levels of care: (1) intensive, (2) high-dependency and (3) special care.[6] Following the location analysis of birth centres, the same methodology is applied to analyse the optimal location of NICUs.

TABLE 6 Regional projections of the population of females aged 15–39 years

Region	Year 2014	2019	2024	2029	2034	2039
Female population aged 15–39 years (000s) (n)						
East	911	924	933	945	954	978
East Midlands	716	727	732	743	744	758
London	1707	1770	1778	1793	1824	1881
North East	408	409	407	408	404	407
North West	1127	1126	1118	1118	1109	1121
South East	1352	1359	1363	1382	1389	1421
South West	786	796	800	813	818	837
West Midlands	901	916	923	934	937	955
Yorkshire and the Humber	855	863	863	869	866	876
England	8763	8890	8918	9004	9044	9234
Total	17,525	17,779	17,836	18,009	18,088	18,467
Change from 2014 (%)						
East	0.0	+1.5	+2.4	+3.8	+4.7	+7.4
East Midlands	0.0	+1.4	+2.2	+3.7	+3.9	+5.8
London	0.0	+3.7	+4.2	+5.1	+6.9	+10.2
North East	0.0	+0.3	–0.2	+0.0	–1.0	–0.3
North West	0.0	+0.0	–0.7	–0.8	–1.6	–0.5
South East	0.0	+0.5	+0.8	+2.2	+2.7	+5.1
South West	0.0	+1.3	+1.9	+3.5	+4.2	+6.5
West Midlands	0.0	+1.6	+2.4	+3.6	+4.0	+5.9
Yorkshire and the Humber	0.0	+1.0	+0.9	+1.6	+1.3	+2.5
England	0.0	+1.4	+1.8	+2.8	+3.2	+5.4
Total	0.0	+1.4	+1.8	+2.8	+3.2	+5.4

Note
Data from the Office for National Statistics.[37]

Data

Very low-birthweight infants

The number of VLBW infants (weighing < 1500 g) per year was computed by regression analysis for each LSOA using the complete records of the BadgerNet database of NHS England (see *Chapter 4*, *Neonatal data*).

Other data

Information submitted by the NDAU for the 2015 National Neonatal Audit Programme report[1] provided the locations of 161 existing neonatal care units, containing 45 ICUs.

Travel times for patients between the LSOAs of England (country divisions with similar population sizes as defined in 2011) and the 161 existing neonatal units were estimated (see *Chapter 4*, *Geographic areas*).

The map of England was provided by OpenStreetMap© (see *Figures 8–11*).[60]

Model

To study the location of NICUs in England, we adapted the model criteria presented in *Decision criteria* to focus on improving the clinical outcome for VLBW infants. To do so, we aimed to include ≥ 100 VLBW infants per year in all modelled NICUs. As a result, the set of optimised criteria is a combination of travel time criteria and VLBW number criteria, as detailed in *Table 5*.

Estimation

In the first stage, the location analysis was restricted to the $H = 45$ existing NICUs. In the second stage, the location optimisation was extended to all $H = 161$ existing neonatal care locations.

For both stages, the MOO based on the NSGA-II method[56] and the location model (see *Model*) was applied to the data described in *Data*, similar to the computation of optimal maternity locations (see *Maternity unit location analysis*).

The population size was set to $P = 100$ and remained constant through generations. The population was propagated through $G = 200$ generations; this value would provide a steady state, as described in *Convergence metrics*.

The MOO process was run independently 10 times for all $h \in [\![1; H]\!]$ with different first generations. Such iterations improve the reliability of analysing the content of the Pareto front configurations.

Results

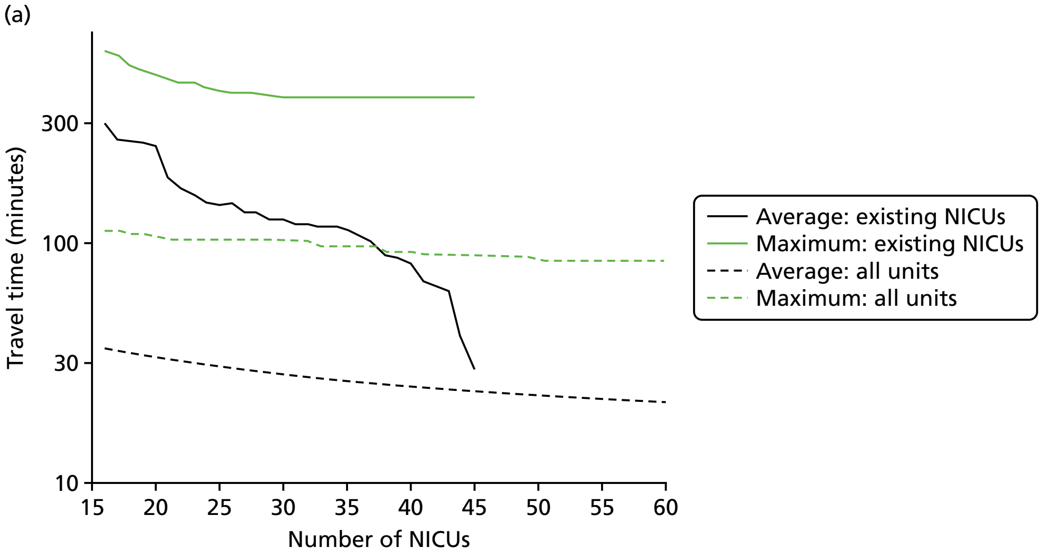
The analysis of the Pareto front configurations provided by the optimisation process enlightens the relationship between the number of NICUs in England, the access to neonatal intensive care and the size of units.

Access to care

In *Figure 7a*, the best achievable average and maximum travel times from mother to unit both decrease as the number of NICUs increases. This relationship is tenuous, as the best achievable maximum travel time remains constant in the case of existing NICUs and it decreases by only 5 minutes between 45 and 60 units in the case of all possible locations. This can be explained: the optimisation criteria include the average travel time; as a result, Pareto front configurations are optimal for a majority of mothers. Such comparison highlights that the maximum travel time from any mother to the closest unit could be improved significantly by changing the location of units, reducing the maximum travel time from 142 minutes to 86 minutes with the current number of 45 NICUs.

Size of units

Figure 7b shows the relationship between the number of NICUs and the number of VLBW infant admissions in Pareto front configurations. As expected, the number of admissions decreases as the number of NICUs increases, with very similar figures for both cases based on existing NICUs or all possible locations between 16 and 36 units. Based on the discovered results, the largest configuration with a minimum number of admissions of ≥ 100 VLBW infants contains 36 NICUs if only selecting existing units, and up to 48 NICUs if using all potential locations. Hence, it is possible to raise the minimum number of admissions from 29 to 100, while keeping the current number of 45 NICUs by changing their location.



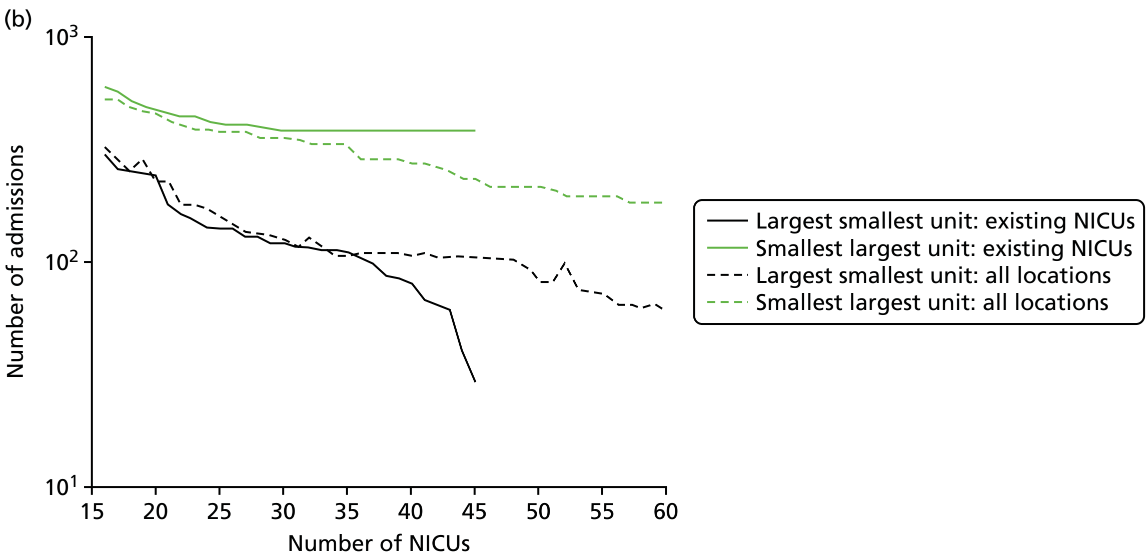


FIGURE 7 Influence of the number of NICUs on the access to care. (a) Travel time (minutes) (configuration: average and maximum travel times); (b) VLBW admissions (configuration: minimum and maximum number of VLBW admissions per year); (c) proportion of VLBW infants in target (configuration: proportion of admissions within 30 minutes of mother's home, proportion of VLBW admissions in a unit with ≥ 100 VLBW admissions per year, and proportion of VLBW infants in a unit with ≥ 100 VLBW admissions per year and within 30 minutes of home); and (d) proportion of VLBW infants in target [same configuration as (c) but only showing the best performing configurations]. Quantified by the travel time to units and the proportion of women in target time, on the number of admissions and on the proportion of births occuring in large units. Dotted lines show configurations in which NICUs can be chosen from any neonatal care location; solid lines show configurations in which choice of location is limited to current NICU locations. (*continued*)

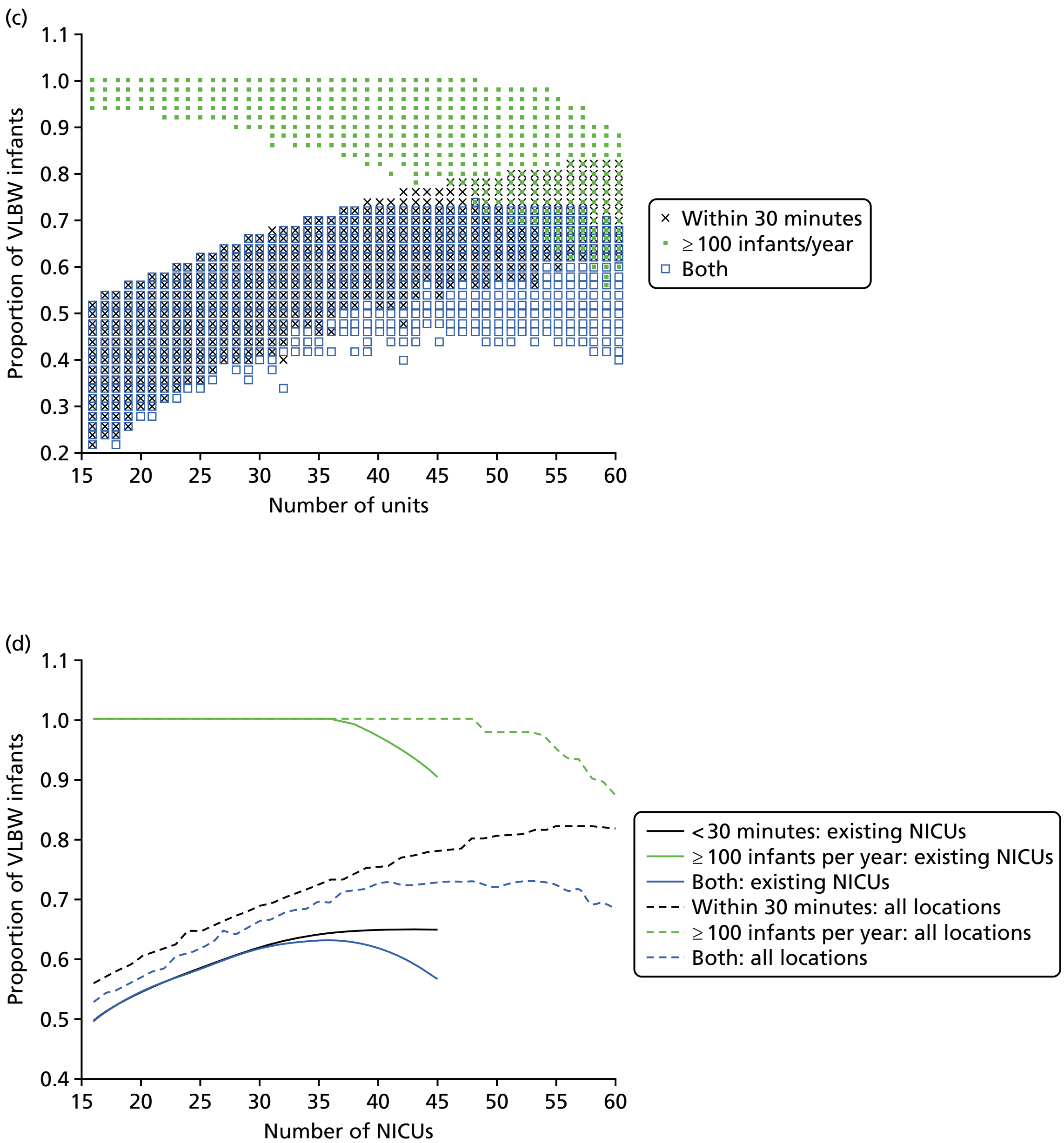


FIGURE 7 Influence of the number of NICUs on the access to care. (a) Travel time (minutes) (configuration: average and maximum travel times); (b) VLBW admissions (configuration: minimum and maximum number of VLBW admissions per year); (c) proportion of VLBW infants in target (configuration: proportion of admissions within 30 minutes of mother's home, proportion of VLBW admissions in a unit with ≥ 100 VLBW admissions per year, and proportion of VLBW infants in a unit with ≥ 100 VLBW admissions per year and within 30 minutes of home); and (d) proportion of VLBW infants in target [same configuration as (c) but only showing the best performing configurations]. Quantified by the travel time to units and the proportion of women in target time, on the number of admissions and on the proportion of births occuring in large units. Dotted lines show configurations in which NICUs can be chosen from any neonatal care location; solid lines show configurations in which choice of location is limited to current NICU locations.

Compromise

The impact of the NICU national configuration on the access of care and clinical outcome can be summarised in criteria 3, 4 and 1 (see *Decision criteria*). Such measures are based on the proportion of mothers living within 30 minutes of the closest NICU, and the proportion of VLBW infants being admitted to NICUs with ≥ 100 VLBW infants per year. *Figure 7c* shows the criterion values of all Pareto front configurations

Figure 7d compares the benefit of changing the number of NICUs using only existing NICUs or all potential locations.

Based on the estimated results, the maximum number of NICUs needed to have 100% of VLBW infants attending large units is 36 with existing NICUs and 48 with all potential locations. This observation is consistent with the analysis shown in *Figure 7b*. In particular, the proportion of VLBW infants attending large units can be increased from 90% to 100% while keeping the current number of 45 NICUs by changing their location.

Moreover, the proportion of mothers within 30 minutes of the closest unit can be improved by a minimum of 7% by releasing the set of NICUs from existing locations to all potential locations. In particular, the proportion can be increased from 65% to 78% while keeping the current number of 45 NICUs.

Finally, the proportion of patients meeting both targets (being within 30 minutes of a NICU and for the NICU to admit ≥ 100 VLBW infants per year) can be increased from 56% to 73% while keeping 45 NICUs.

Example of an alternative neonatal intensive care unit configuration

Going beyond the performance metrics, it is interesting to study what the discovered Pareto front configurations mean for the patients locally. To do so, an example of an alternative NICU configuration was selected using the following method:

1. Select the Pareto front configuration in the highest quartile of the proportion of patients both attending units with ≥ 100 VLBW infants per year and living within 30 minutes of the closest unit. This allows the selection of the best-performing configurations.
2. Select the Pareto front configurations with all patients attending units with ≥ 100 VLBW infants per year.
3. Select the Pareto front configurations with the highest number of units.

From the 88,800 discovered configurations, the selection led to two configurations with 48 units, of which the one that was closest to the current configuration was chosen.

In *Table 7*, the criterion values of this alternative configuration are compared with the model of the current state regarding the care of VLBW infants. The admission numbers are also provided for the infants receiving > 24 hours of intensive care (in an ICU), regardless of their weight.

Overall, the alternative configuration, shown in *Figure 8*, offers a more homogeneous set of units in terms of admissions. Indeed, although the number of VLBW infant admissions, if VLBW infants attend their closest NICU, varies from 29 to 450 per year in the current model, it varies from 101 to 241 per year in the alternative configuration. The latter has the advantage of increasing the proportion of patients attending units with ≥ 100 VLBW infants per year from 90% to 100% while keeping a similar number of units. Furthermore, the proportion of mothers within 30 minutes of the closest unit is raised from 65% to 73%.

For instance, the area including Somerset, Bristol, Wiltshire and Gloucestershire, in the alternative configuration, is covered by one NICU in Bristol (with 143 VLBW infants), one NICU in Bath (with a predicted admission of 101 VLBW infants per year) and one NICU in Gloucester (with 139 VLBW infants), whereas it is currently covered by two NICUs in Bristol (with a predicted admission of 149 and 178 VLBW infants per year) if VLBW infants attend their closest NICU. This local alternative configuration offers faster access to the population outside Bristol, whereas the impact on the Bristol population is limited.

In the current model (assuming that VLBW infants attend their closest NICU), the Greater London area (as shown in *Figure 9*) is covered by Chelsea and Westminster Hospital (with a predicted admission of 32 VLBW infants per year), St Thomas' Hospital (with a predicted admission of 29 VLBW infants per year), Homerton (with a predicted admission of 450 VLBW infants per year), King's College (with a predicted admission of 136 VLBW infants per year), Queen Charlotte's (with a predicted admission of 256 VLBW infants per year), Royal London Hospital (with a predicted admission of 210 VLBW infants per year), St George's (with a predicted admission of 162 VLBW infants per year), St Peter's (with a predicted admission of 379 VLBW infants per year) and University College London (with a predicted admission of 77 VLBW infants per year). In the alternative configuration, intensive care would be provided by Barnet Hospital (with a predicted admission of 127 VLBW infants per year), Chelsea and Westminster (with a predicted admission of 155 VLBW infants per year), North Middlesex University Hospital in Edmonton (with a predicted admission of 241 VLBW infants per year), Newham General Hospital (with a predicted admission of 140 VLBW infants per year), Northwick Park (with a predicted admission of 159 VLBW infants per year), Queen Elizabeth Hospital in Woolwich (with a predicted admission of 117 VLBW infants per year), Royal London (with a predicted admission of 153 VLBW infants per year), St Helier Hospital (with a predicted admission of 191 VLBW infants per year), Kingston upon Thames (with a predicted admission of 119 VLBW infants per year) as well as Wexham Park Hospital in Slough (with a predicted admission of 124 VLBW infants per year) and Darrent Valley Hospital in Dartford (with a predicted admission of 131 VLBW infants per year) on the periphery of Greater London. The number of VLBW infants per year would vary from 117 to 241 in Greater London as opposed to 29 to 379 with the current mapping and VLBW infants attending their closest NICU. As a result, all patients in Greater London could attend a unit with ≥ 100 VLBW infants per year. Moreover, as can be seen in *Figures 9–11* and *Table 7*, the LSOAs in which mothers can reach a NICU within 30 minutes would be extended from central London to the peripheral areas (see *Appendix 1*).

(a)

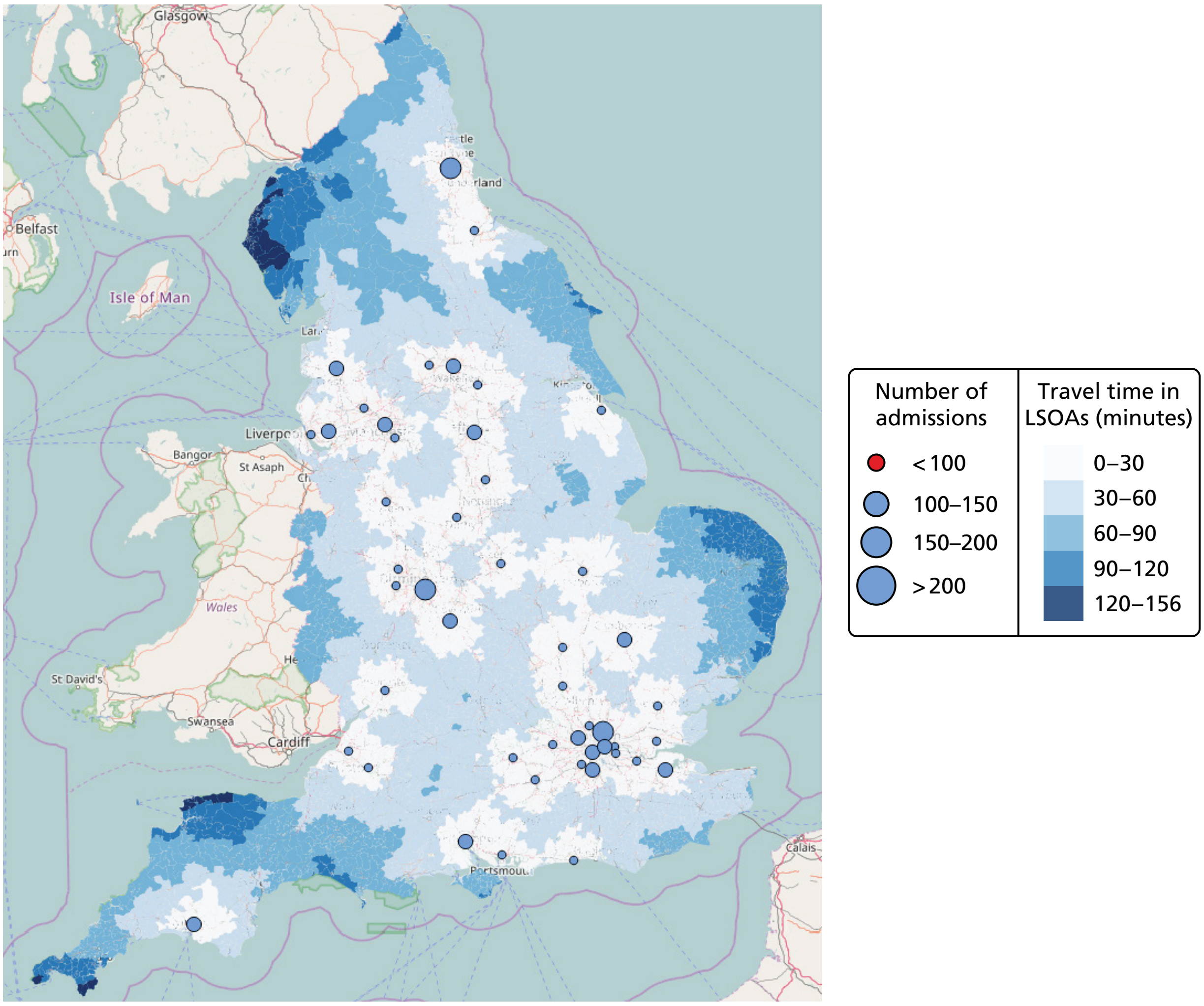


FIGURE 8 Example of optimal configuration in England with 48 NICUs. (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); (b) number of admissions per year by unit location; and (c) key performance indicators. (*continued*)

(b)

(c)

Average travel time	26 minutes
Maximum travel time	143 minutes
Mothers within 30 minutes of closest NICU	73%
VLBW infants attending unit with ≥ 100 VLBW infants per year	100%
Minimum number of VLBW admissions	101
Maximum number of VLBW admissions	241
Mothers and VLBW infants within 30 minutes of closest NICU and attending a unit with ≥ 100 VLBW infants per year	73%

FIGURE 8 Example of optimal configuration in England with 48 NICUs. (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); (b) number of admissions per year by unit location; and (c) key performance indicators.

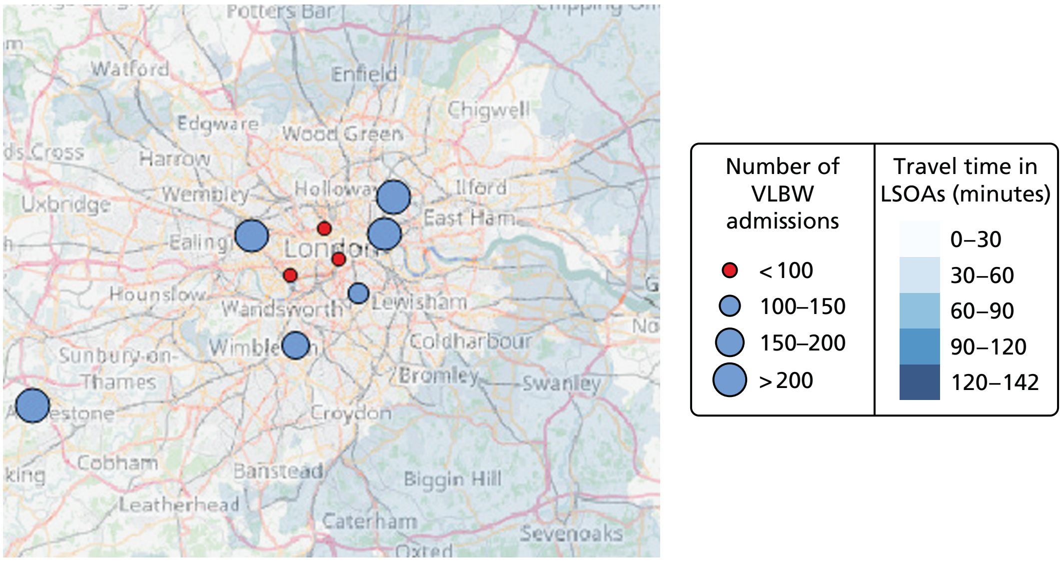


FIGURE 9 Example current configurations for neonatal units with a focus on the Greater London area, with 45 NICUs in England. Admission numbers assume that the closest appropriate unit is used. Chelsea and Westminster (32 VLBW infants), Guy's and St Thomas' (29 VLBW infants), Homerton (450 VLBW infants), King's College (136 VLBW infants), Queen Charlotte's (256 VLBW infants), Royal London Hospital (210 VLBW infants), St George's (162 VLBW infants), St Peter's (379 VLBW infants) and University College London (77 VLBW infants). © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright).

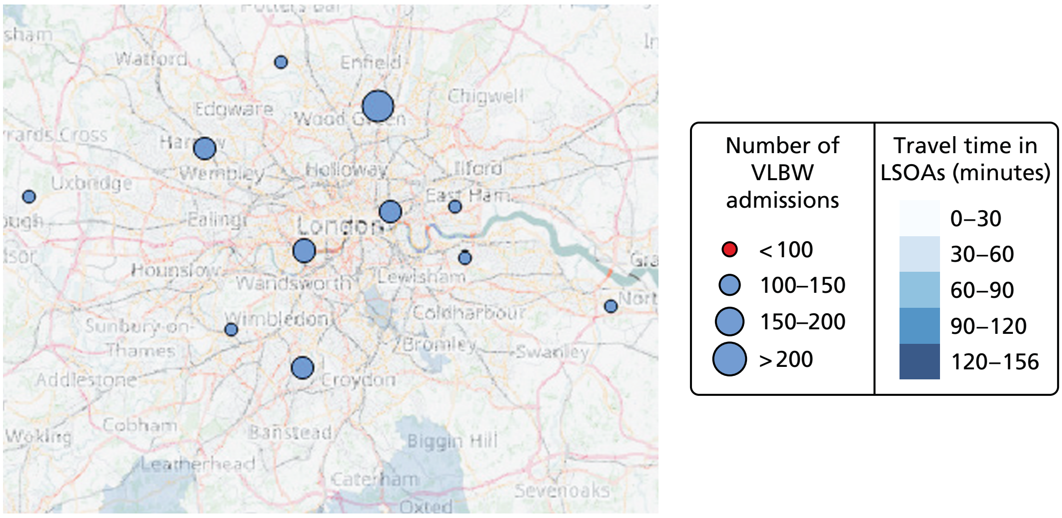


FIGURE 10 Example alternative configuration for neonatal units in the Greater London area, with 48 NICUs in England. Admission numbers assume that the closest appropriate unit is used. Barnet (127 VLBW infants), Chelsea and Westminster (155 VLBW infants), Edmonton (241 VLBW infants), Newham (140 VLBW infants), Northwick Park (159 VLBW infants), Queen Elizabeth – Woolwich (117 VLBW infants), Royal London (153 VLBW infants), St Helier (191 VLBW infants), Kingston upon Thames (119 VLBW infants), Wexham Park – Slough (124 VLBW infants) and Dartford (131 VLBW infants). © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright).

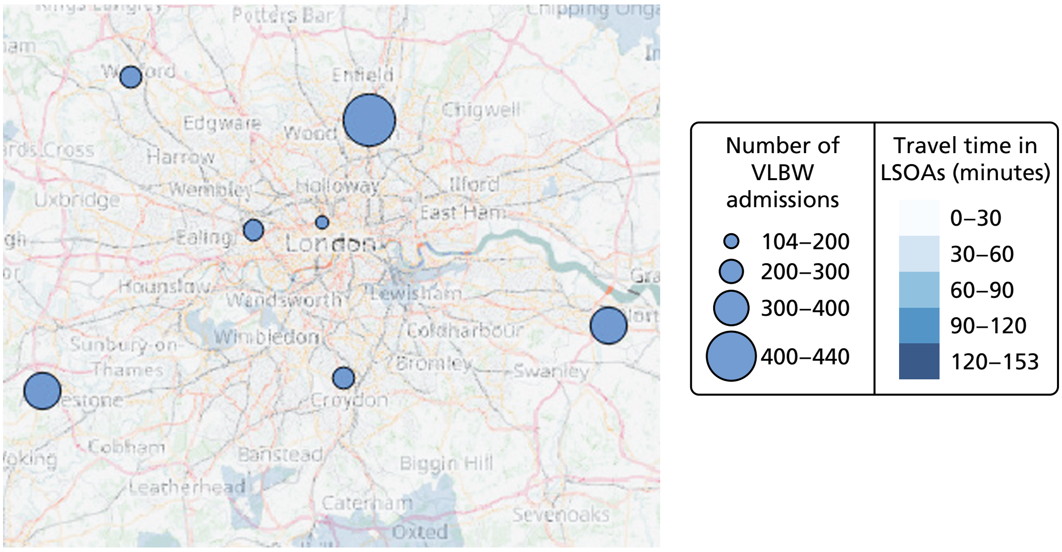


FIGURE 11 Example centralised configuration for neonatal units in the Greater London area, with 30 NICUs in England. Admission numbers assume that the closest appropriate unit is used. Queen Charlotte's (231 VLBW infants), Croydon (238 VLBW infants), Dartford (343 VLBW infants), St Peter's (356 VLBW infants), Edmonton (440 VLBW infants), University College London (176 VLBW infants) and Watford (214 VLBW infants). © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright).

TABLE 7 Comparison of the performances of two potential configurations with the current state, at the three levels of care

Patients (by care level)	Average travel time (minutes)	Maximum travel time (minutes)	% (of infants) within 30 minutes	Minimum number of admissions	Maximum number of admissions	% (of infants) in units with ≥ 100 VLBW infants per year
Current configuration: 45 NICUs + 78 LNUs + 38 SCUs (model)						
VLBW	28	142	65	29	450	90
ICU				50	779	
HD	17	131	90	31	322	N/A
SC	14	82	95	127	965	N/A
Example of centralised configuration: 30 NICUs + 30 LNUs + 30 SCUs						
VLBW	29	153	64	104	440	100
ICU				108	762	
HD	2	99	82	96	610	N/A
SC	17	89	89	137	1984	N/A
Example of alternative configuration: 48 NICUs + 78 LNUs + 35 SCUs						
VLBW	26	142	73	101	241	100
ICU				175	417	
HD	16	94	92	31	237	N/A
SC	Identical to current configuration					

HD, high dependency; N/A, not applicable; SC, special care.

A caveat of the observations around London is that we have used estimated road travel times to choose between hospitals. It is possible that the order of choice may be different if public transport is used (as is common in London). However, the observation remains that the hospitals in Greater London are currently concentrated in the centre of London despite many mothers living in the more outlying regions.

Example of a centralised neonatal intensive care unit configuration

An example of a centralised NICU configuration with 30 units was selected using the following method:

1. Select the Pareto front configurations with 30 units.
2. Select the Pareto front configurations in the best half of the proportion of patients both attending units with ≥ 100 VLBW infants per year and living within 30 minutes of the closest unit.
3. Select the Pareto front configurations with all patients attending units with ≥ 100 VLBW infants per year.
4. Select the Pareto front configurations with the highest number of existing NICUs.

If reducing the number of NICUs to 30 optimal locations, the impact on the access of care would be limited as the average travel time would increase to 29 minutes (+1 minute), the maximal travel time would increase to 153 minutes (+11 minutes) and the proportion of mothers within 30 minutes of the closest unit would decrease to 64% (–1%). The main impact would be focused on the minimum number of admissions, which would increase from 29 to 104 VLBW infants per year. Furthermore, changing the location of NICUs by upgrading existing LNUs would equalise the distribution of units in the country, which would then reduce the maximum number of VLBW infant admissions from 450 to 440 per year.

Building a resilient network

Definition of the resilience probability

In order to analyse the possible scenarios, it is interesting to highlight which locations are more likely to appear in the optimal configurations. To do so, we compute the resilience probability of every location in accordance with the following method:

1. Select the Pareto front configurations with 35 to 55 units (current number 45 ± 10). This allows the selection of configurations close to the current state.
2. Select the Pareto front configurations in the highest quartile of the proportion of patients both attending units with ≥ 100 VLBW infants per year and living within 30 minutes of the closest unit. This allows us to select the best-performing configurations.
3. Compute the probability $p(u_i|h)$ of each unit u_i to appear in the selected '*h*-configurations' (configurations with *h* units):

$$p(u_i|h) = \frac{Number\ of\ h\text{-}configurations\ with\ u_i}{h \times number\ of\ h\text{-}configurations}. \tag{3}$$

Such probability verifies:

$$\forall h \textstyle\sum_{i=1}^{H} p(u_i|h) = 1. \tag{4}$$

1. Compute the probability $p(u_i)$ of each unit u_i to appear in the selected configurations:

$$p(u_i) = \frac{1}{(h_{max} - h_{min} + 1)} \textstyle\sum_{h=h_{min}}^{h_{max}} p(u_i|h). \tag{5}$$

Such probability verifies:

$$\textstyle\sum_{i=1}^{H} p(u_i) = 1. \tag{6}$$

The locations that appear the most often in the optimal configurations will have a higher resilience probability. Such an indicator is useful in order to select the locations that are the most resilient to network changes.

Resilience charts

Figure 12 shows the resilience or optimality probability of the locations of four Operational Delivery Networks (ODNs), as well as their existing level of care. The equivalent charts for other ODNs are available in *Appendix 1*. If the resilience probability is high, then the location is likely to contribute to building a NICU network resilient to changes. For instance, the unit in Dartford has a very high resilience probability, which may be attributable to the hospital being easily accessible by road. The unit in Plymouth also has a high resilience probability because there is a high demand in the local area. It is interesting to compare the probability graph with the existing level of care. Some units with a high resilience probability, such as Dartford, Haywards Heath and Gloucester, do not currently provide intensive care. In areas such as the South London Neonatal ODN, the resilience probability is rather low because several units are present in the restricted geographic area. As a consequence, selecting one unit or another would not significantly change the access performances.

Neonatal high-dependency care location analysis

High-dependency care is provided by both NICUs and LNUs (see *Chapter 1*, *Clinical importance*). NICU locations have been previously selected (see *Figure 8*), and, here, LNUs are added to study the coverage of more units providing high-dependency care.

Data

The demand for high-dependency care was modelled as the number of neonatal admissions needing > 24 hours of high-dependency care. It was estimated in all LSOAs as the number of neonatal admissions predicted by the regression analysis (see *Chapter 4*, *Geographic data coverage and predicting neonatal demand*) and multiplied by a factor of 0.185.

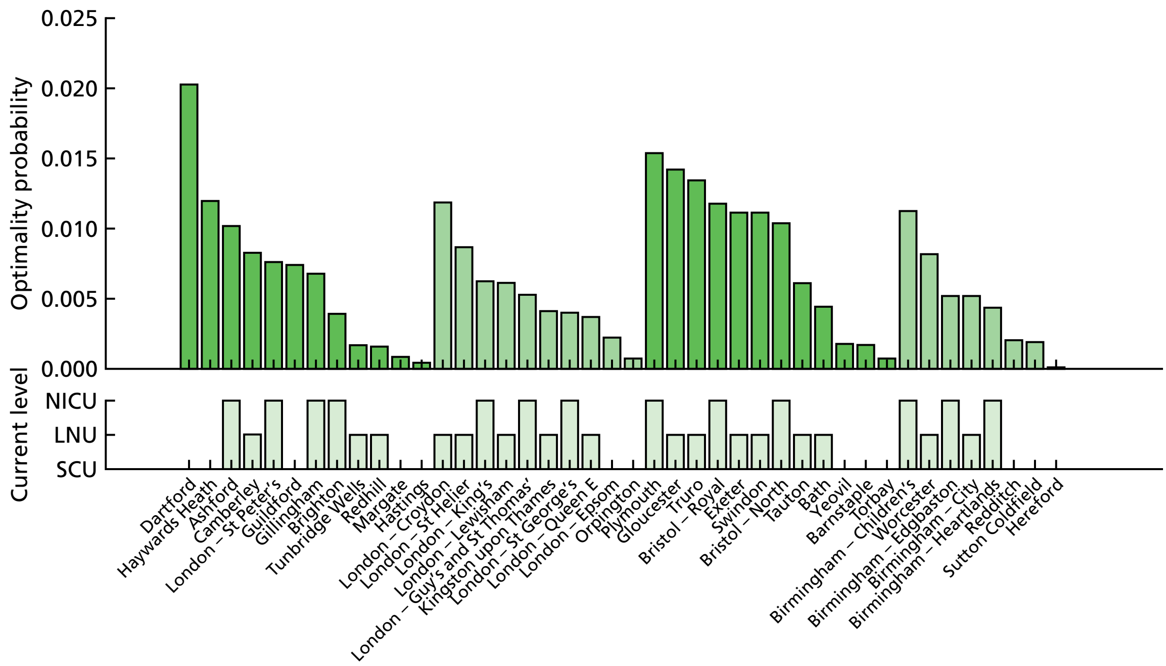


FIGURE 12 Optimality probability and existing level of neonatal care of locations in South East England. Neonatal ODN (first dark green group), South London Neonatal ODN (first medium green group), South West Neonatal ODN (second dark green group) and Southern West Midlands Maternity and Newborn Network (second medium green group). For a full explanation of this chart, see *Appendix 1*.

The lists of existing neonatal units, geographic data and travel times were the same as those used in the intensive care location analysis (see *Neonatal intensive care location analysis*).

Example of an alternative local neonatal unit configuration

Following the example of an alternative NICU configuration in *Neonatal intensive care location analysis*, this section focuses on selecting 78 LNU locations to build an optimal high-dependency network. The number 78 corresponds with the current number of LNUs in England.

Estimation

To study the location of LNUs (providing high-dependency and special care) in England, the model criteria presented in *Decision criteria* were adapted to become a combination of travel time criteria and high-dependency number criteria, as detailed in *Table 6*. The 48 NICUs selected in the previous example of centralised NICU configuration (see *Neonatal intensive care location analysis*, *Example of a centralised neonatal intensive care unit configuration*) were fixed and the 113 remaining units were considered as potential LNUs.

In order to find 78 optimal LNU sites, the optimisation was applied to the high-dependency demand (see *Neonatal high-dependency care location analysis*, *Data*) and the 131 potential locations, using the NSGA-II method[56] and the defined criteria, with the same population and generation parameters than for the computation of optimal NICUs locations (see *Neonatal high-dependency care location analysis*, *Data*). The MOO process was run independently 10 times with different first generations, leading to 1000 Pareto front configurations of 78 locations.

Selection

An example of a centralised LNU configuration with 78 units was selected using the following method:

1. Select the best half of the Pareto front configurations in accordance with the proportion of patients living within 30 minutes of the closest unit.
2. Select the best half of the Pareto front configurations in accordance with maximum travel time.
3. Select the best half of the Pareto front configurations in accordance with the maximum number of admissions.
4. Select the best half of the Pareto front configurations in accordance with the minimum number of admissions.
5. Select the Pareto front configurations with the highest number of existing LNUs or NICUs.

Discussion

The map and list of units of the selected configuration are available in *Appendix 1*. The performances can be compared with the current state model in *Figure 9*. As the total number of units providing high-dependency care remains similar (126 instead of 123), the average travel time, the proportion of mothers within 30 minutes of the nearest unit and the minimum number of high-dependency admissions remains almost unchanged in comparison with the current state model. However, the maximum travel time could be reduced to 94 minutes (–37 minutes) by changing the location of units; this is made possible by selecting NICUs and LNUs from all existing care locations. Moreover, the units would be more homogeneous as the maximum number of high-dependency admissions would be reduced to 241 (–85).

Example of a centralised local neonatal unit configuration

Following the example of a centralised NICU configuration (see *Neonatal intensive care location analysis*, *Example of a centralised neonatal intensive care unit configuration*), this section focuses on selecting 30 LNU locations to build an optimal centralised LNU network.

Estimation

The 30 NICUs selected in the previous example of centralised NICU configuration (see *Neonatal intensive care location analysis*, *Example of a centralised neonatal intensive care unit configuration*) were fixed and the 131 remaining units were considered as potential LNUs. The MOO process was run independently 10 times with different first generations, leading to 1000 Pareto front configurations of 30 locations.

An example centralised LNU configuration with 30 units was selected using the same method as described in *Neonatal high-dependency care location analysis*, *Selection*.

Discussion

The map and list of units of the selected centralised LNU configuration are available in *Appendix 1*. The performances can be compared with the current state model in *Figure 9*. By reducing the number of high-dependency care units from 123 (45 NICUs and 48 LNUs) to 60 (30 NICUs and 30 LNUs), the quality of access would deteriorate slightly as the average travel time would increase to 22 minutes (+5 minutes) and the proportion of mothers within 30 minutes of the nearest unit would decrease to 82% (–8%). However, such changes would be minimal considering that the number of units providing high-dependency care would be cut in half. It is interesting to notice that the maximum travel time could be reduced to 99 minutes (–34 minutes) by changing the location of units.

Neonatal special care location analysis

The special care is provided by all neonatal care units (see *Chapter 1*, *Clinical importance*). NICU and LNU locations have previously been selected, and here SCUs are added to study the coverage of providing special care.

Data

The demand for special care was modelled as the number of neonatal admissions requiring > 24 hours of special care. It was estimated in all LSOAs as the number of neonatal admissions predicted by the regression analysis (see *Chapter 4*, *Geographic data coverage and predicting neonatal demand*) and multiplied by a factor of 0.825.

The lists of existing neonatal units, geographic data and travel times were the same as those used in the intensive care location analysis (see *Neonatal intensive care location analysis*, *Data*).

Example of a centralised special care unit configuration

Estimation

To study the location of SCUs in England, the model criteria presented in *Decision criteria* were adapted to become a combination of travel time criteria and high-dependency number criteria, as detailed in *Table 6*.

The 30 NICUs and 30 LNUs selected in the previous examples (see *Neonatal intensive care location analysis*, *Example of a centralised neonatal intensive care unit configuration*, and *Neonatal high-dependency care location analysis*, *Example of a centralised neonatal intensive care unit configuration*) were fixed and the 101 remaining units were considered as potential SCUs.

In order to find 30 optimal SCU sites, the optimisation was applied to the special care demand (see *Neonatal high-dependency care location analysis*, *Data*) and the 101 potential locations, using the NSGA-II method[56] and the defined criteria, with the same population and generation parameters as for the computation of optimal NICU locations (see *Neonatal intensive care location analysis*, *Estimation*).

The MOO process was run independently 10 times with different first generations, leading to 1000 Pareto front configurations of 30 locations. An example of a centralised configuration with 30 SCUs was selected using the same method (see *Neonatal high-dependency care location analysis*, *Selection*).

Discussion

With 90 optimal units providing special care, instead of 161, the quality of access to care would be only slightly deteriorated, with an average travel time of 17 minutes (+3 minutes), a maximum travel time of 89 minutes (+7 minutes) and 89% (–6%) of mothers within 30 minutes of the closest hospital. The main impact would be on the increased number of special care admissions, ranging from 137 (+10) to 1984 (+1019).

Location analysis discussion

Maternity units

For both maternity care and neonatal care, choosing an optimal configuration of units is a compromise between fair access to local care for every patient and access to experienced medical staff in units with a threshold number of patients.

The Royal College of Obstetricians and Gynaecologists recommends that all maternity units should ideally receive ≥ 6000 babies per year in order to guarantee the 24/7 (24 hours a day, 7 days a week) presence of a consultant on site.[61] The maternity location analysis (see *Maternity unit location analysis*) has shown that in order to achieve 100% of births occurring in such units, the number of birth centres would need to be reduced from 161 to approximately 72. Although such change is unrealistic, reducing the number of birth centres from 161 to 140 optimal locations could increase the proportion of births in 24/7 units from 30% to 50%.

Reducing the size of the network by a small number of units does not significantly change the average travel time if the new national configuration is optimally chosen, but it increases the maximal travel time. Average travel times have the potential to hide large effects that only affect a few people in more remote areas.

Access to local care is indicated by the proportion of patients within 30 minutes of the closest unit. Achieving this criterion conflicts with achieving all births in 24/7 consultant-led units. By reducing the number of birth centres from 161 to approximately 65, the proportion of patients meeting both the travel time target and the unit size target would be increased from 24% to 82%: the maximum achievable based on our results. Such a low number of birth centres is not necessarily ideal, but it must be expected that, as the number either increases or reduces, the proportion of patients meeting both targets will reduce either because distances become too large or because units become too few.

Neonatal units

An alternative neonatal configuration was analysed in *Neonatal intensive care location analysis*, *Example of a centralised neonatal intensive care unit configuration*. By changing the level of care provided by some units, but not the total number of units, it should be possible to admit all VLBW infants to units with ≥ 100 VLBW infants per year. This configuration would guarantee access to experienced staff to all patients. The neonatal units would also be more homogeneous in size, with fewer units receiving a very low or a very high number of VLBW infants (or other infants requiring intensive or high-dependency care). Note that the nurse capacity needed at the national scale for this configuration would remain similar to the current capacity, as the number of units for each level of care would remain almost identical; reconfiguration of this type would therefore be about improving care and access for patients rather than reducing costs to service providers. These results show that the current location of NICUs is not optimal for either access (closeness) or optimising the use of large NICUs admitting ≥ 100 VLBW infants per year; it is theoretically possible to improve both access to and coverage of large NICUs simultaneously if there was the opportunity to upgrade some units and downgrade others.

The analysis of the Greater London area has shown that current NICUs are concentrated in the city centre, probably for historical reasons. With the alternative configuration, the optimal locations for NICUs would be more evenly spread in neonatal units in the conurbation, closer to residential areas and main road axes. The analysis is similar in the Bristol area, where the local population would benefit from NICUs in Bath (rather than two NICUs in Bristol, the current configuration). It is likely that this observation is transferable to other conurbations, such as Birmingham and Manchester.

London is likely to be somewhat of a special case because of the greater provision and use of public transport than in other regions. We have estimated travel times based on typical road travel times. A more focused study of London is likely to benefit from use of public transport times in addition to road distances.

Nevertheless, the finding that hospitals in London are not focused on the location where parents live is likely to still be significant; access to care is likely to improve by some movement of locations of care away from central London towards the population centres further away from the city centre.

The interface between obstetric and neonatal care

In our work, we have modelled maternity and neonatal care as two separate problem spaces. For the large majority of births and infants, there is no need for rapid access to specialist neonatal care; however, for a proportion of infants, rapid access to specialist neonatal care may prevent loss of life or avoid/reduce disability. Outcomes for extremely premature infants are improved for those infants born in hospitals with high-volume specialist neonatal care units.[19] Blondel *et al.*[62] reported that in 2003, across a range of European regions, 2–28% of birth units were associated with a 'large neonatal unit' (≥ 50 admissions per year for infants with a gestational age of < 32 weeks). Between 8% and 61% of all births were in hospitals with large neonatal units, but 37–76% of births of infants with a gestational age of < 32 weeks were achieved in such units (20–54% of births of infants with a gestational age of < 32 weeks were achieved in such units without requiring in utero transfer).

There are three general models for trying to achieve births of preterm infants in units with large neonatal units (and these models may be mixed): (1) consolidation of births into higher-volume units that may sustain a large neonatal unit, (2) identifying mothers at a high risk of preterm delivery and booking those mothers to deliver in high-volume neonatal units and (3) in utero transfer during labour. Blondel *et al.*[62] noted that different European regions were achieving reasonable rates of birth of premature infants in units with large neonatal units by using different organisational models; however, there was still a significant proportion of births (24–63%) of premature infants not occurring in units with large neonatal units.

Increased consolidation of childbirth could increase the proportion of preterm births in units with large neonatal units (especially if the large neonatal units were optimally located to be better matched to demand). But, as Blondel *et al.*[62] noted, organisation of services around these high-risk births may not be acceptable to the larger population of low-risk mothers whose infants will not require any specialist obstetric neonatal care, and for whom birth is not a medical procedure.

Even given the complexities of balancing the wishes of low-risk mothers and the needs of high-risk mothers, co-ordinated planning of birth and neonatal care could potentially improve services for all. In this study, we have not explicitly linked birth and neonatal care models, but we believe that this would be valuable future work.

General comments

Resilience of solutions

Given the high number of possible configurations, it is impossible to compute exactly the best performances and it is time-consuming to compare lots of scenarios. In the cases of the centralised and the alternative scenarios, a selection method has been proposed to extract a few options from thousands of possibilities, using the performance criteria. It is also possible to choose the potential scenarios that are the closest to the current state (in order to find more pragmatic solutions that require fewer changes to the existing configuration).

Analysing scenarios with up to 161 locations is a complex task. To do so, the resilience probability defined in *Building a resilient network* measures the optimality of locations for a given set of optimisation criteria. As the national neonatal care network is constantly changing, the resilience probability is an interesting way to study which locations are likely to build an optimal network, adaptable to the demand in the long term. The charts presented (see *Resilience charts* and *Appendix 1*) show a picture of the resilience at the national level in order to build a NICU network. The locations with the highest resilience probability should be considered in priority in the case of a network restructuring as these locations are generally less sensitive to changes elsewhere in the network. Such methodology is easily transferable to the other levels of care, and more widely to any geographic analysis.

Networks

The neonatal units are organised by ODNs. The model presented in *Neonatal intensive care location analysis*, *Model*, does not constrain the optimisation of units in the ODN framework, for instance with a minimum number of units per network. However, as the optimisation aligns the units with the population, the selected locations are distributed in most networks. The main advantage of this maternity and neonatal location analysis is to consider the delivery network at the national scale rather than at the usual local scale. This is not to say that networks should be avoided, but that networks should be led by, and not lead, choice of locations if reconfiguration is to be designed around patient-centred objectives. Networks are further discussed in the following sections regarding the investigation of their effect in the simulation model.

Limitations of the analysis work

With 161 potential locations to provide neonatal care, there are 1.82×10^{47} possible configurations for maternity and neonatal units. In the case of NICUs, 88,800 Pareto front configurations were discovered after 200 generations of the genetic algorithm, which means that up to 17,760,000 configurations have been analysed. Considering the size of the decision space, it is impossible to be sure that the discovered Pareto front is the best achievable. However, the individual optimisations realised for each number $h \in [\![1; H]\!]$ of units, iterated 10 times with different starting points, lead to consistent results. The smoothness of the graphs in *Figures 2* and *3* allows us to claim that the optimisation processes have reached convergence and, as a result, some local optima.

In this work, we have assumed that care would normally take place in the closest appropriate unit. Where units are close together or where people are on the boundary between two units (and so travel times are not much different between them), this may not always be the case.

Chapter 7 Simulation modelling

Model description

The model assumes a standard direction of flow of infants, from higher levels of care through to lower levels of care, and then exit (*Figure 13*). The infant may enter at any point and may not use all levels of care between the entry point and the exit point. The model assumes that an infant is born in the unit closest to the mother's home location, but is then transferred immediately to the required level of care if the hospital of birth does not provide the necessary level of care.

In the model:

- All surgery-specific care must be within a surgical unit (but the infant may then move to other units for ongoing care).
- All Level 1 (intensive) care of ≥ 48 hours' duration must be in a NICU. Level 1 stays of < 48 hours' duration may also be in a LNU.
- All Level 2 (high-dependency) care of ≥ 48 hours' duration must be in a NICU or LNU. Level 2 stays of < 48 hours' duration may also be in a SCU.

Infant categories and data used in the model

Infants in the model are divided into six categories by gestational age. Infants requiring specialist surgical care form a separate category. Probabilities of movement through the model, and distributions of LOS, depend on the infant category. *Table 8* shows the classification by gestational age and the incidence of deliveries by category.

The model includes a probability of multiple infants per delivery depending on gestational age category at birth (*Table 9*).

Infants have probabilities of entry and transition between levels depending on their gestational age category (*Table 10*). In the model, 'exit' may mean discharge to home, discharge to a non-neonatal unit for ongoing care, or death.

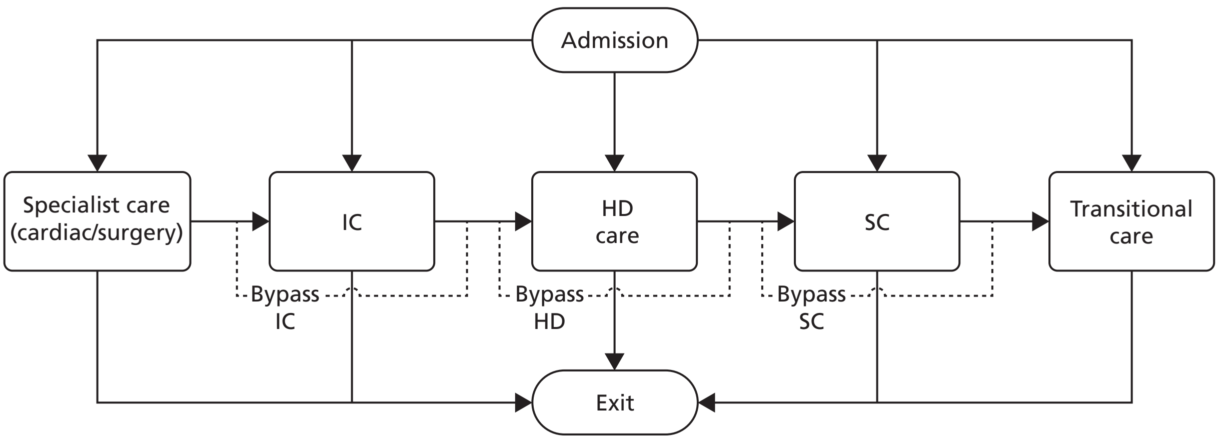


FIGURE 13 Schematic flow of infants through the model. HD, high dependency; IC, intensive care; SC, special care. Any one or more levels may be bypassed: an infant could, theoretically, exit intensive care and directly enter transitional care.

TABLE 8 Infant categories in model (with % of all deliveries not % of infants, as there may be multiple infants per delivery)

Infant category	Gestational age at birth (weeks)	All deliveries (%)	Infants requiring specialist surgical care (%)
1	< 24	0.23	9.71
2	24 to < 27	1.46	11.23
3	27 to < 30	2.89	5.06
4	30 to < 33	6.52	1.87
5	33 to < 36	17.67	0.82
6	≥ 36	71.23	0.93

TABLE 9 Incidence of multiple infants per delivery

	Number of fetuses per delivery (% of deliveries)				
Infant category	1	2	3	4	5
1	83.4184	14.9235	1.6582	0.0000	0.0000
2	85.5238	14.0923	0.3233	0.0606	0.0000
3	85.7055	13.3886	0.8598	0.0461	0.0000
4	84.1907	14.8712	0.9211	0.0170	0.0000
5	86.0862	13.3991	0.5019	0.0088	0.0040
6	97.1603	2.8110	0.0166	0.0069	0.0052

Note
This table shows the percentage of all deliveries with multiple infants.

TABLE 10 Entry and transition probabilities for infants in the model, depending on infant category

		Level moving to (% of patients)					
Entry/exit points	Infant category	Surgical	Level 1	Level 2	Level 3	Level 4	Exit
Entry point	1	0.00	100.00	0.00	0.00	0.00	0.00
	2	0.00	100.00	0.00	0.00	0.00	0.00
	3	0.00	93.41	6.59	0.00	0.00	0.00
	4	0.00	50.34	35.27	14.38	0.00	0.00
	5	0.00	8.77	29.99	59.86	1.38	0.00
	6	0.00	6.32	10.61	73.73	9.34	0.00
	7	100.00	0.00	0.00	0.00	0.00	0.00
Exit surgical	1	0.00	0.00	0.00	0.00	0.00	0.00
	2	0.00	0.00	0.00	0.00	0.00	0.00
	3	0.00	0.00	0.00	0.00	0.00	0.00
	4	0.00	0.00	0.00	0.00	0.00	0.00
	5	0.00	0.00	0.00	0.00	0.00	0.00
	6	0.00	0.00	0.00	0.00	0.00	0.00
	7	0.00	47.37	11.70	39.77	1.17	0.00

TABLE 10 Entry and transition probabilities for infants in the model, depending on infant category (*continued*)

		Level moving to (% of patients)					
Entry/exit points	**Infant category**	**Surgical**	**Level 1**	**Level 2**	**Level 3**	**Level 4**	**Exit**
Exit Level 1	1	0.00	0.00	22.22	0.00	0.00	77.78
	2	0.00	0.00	80.43	0.00	0.00	19.57
	3	0.00	0.00	94.12	0.00	0.00	5.88
	4	0.00	0.00	86.39	6.80	0.00	6.80
	5	0.00	0.00	80.26	17.11	0.00	2.63
	6	0.00	0.00	44.02	47.88	0.00	8.11
	7	0.00	0.00	64.20	11.11	0.00	24.69
Exit Level 2	1	0.00	0.00	0.00	75.00	0.00	25.00
	2	0.00	0.00	0.00	81.08	0.00	18.92
	3	0.00	0.00	0.00	90.70	0.00	9.30
	4	0.00	0.00	0.00	97.83	0.00	2.17
	5	0.00	0.00	0.00	99.07	0.31	0.62
	6	0.00	0.00	0.00	95.26	2.91	1.82
	7	0.00	0.00	0.00	90.00	0.00	10.00
Exit Level 3	1	0.00	0.00	0.00	0.00	4.95	95.05
	2	0.00	0.00	0.00	0.00	15.67	84.33
	3	0.00	0.00	0.00	0.00	24.25	75.75
	4	0.00	0.00	0.00	0.00	27.05	72.95
	5	0.00	0.00	0.00	0.00	22.86	77.14
	6	0.00	0.00	0.00	0.00	11.32	88.68
	7	0.00	0.00	0.00	0.00	20.90	79.10
Exit Level 4	1	0.00	0.00	0.00	0.00	0.00	100.00
	2	0.00	0.00	0.00	0.00	0.00	100.00
	3	0.00	0.00	0.00	0.00	0.00	100.00
	4	0.00	0.00	0.00	0.00	0.00	100.00
	5	0.00	0.00	0.00	0.00	0.00	100.00
	6	0.00	0.00	0.00	0.00	0.00	100.00
	7	0.00	0.00	0.00	0.00	0.00	100.00

Note
Category 7 also includes infants requiring specialist surgical care, regardless of gestational age.

Length of stay

In previous regional work, we identified the log-normal distribution as the best distribution for approximating actual variation in lengths of stay.[33] In this study, we examined whether or not the log transformation was a reasonable distribution to use for the national data set (*Figure 14*). In order to test the distribution across the combined data set, we examined the distribution of lengths of stay when compared with the category-level mean for that category-level group (there are seven infant categories used, and four general levels of care, with one additional level of care for infants requiring surgery, giving 29 category levels; see *Table 10*).

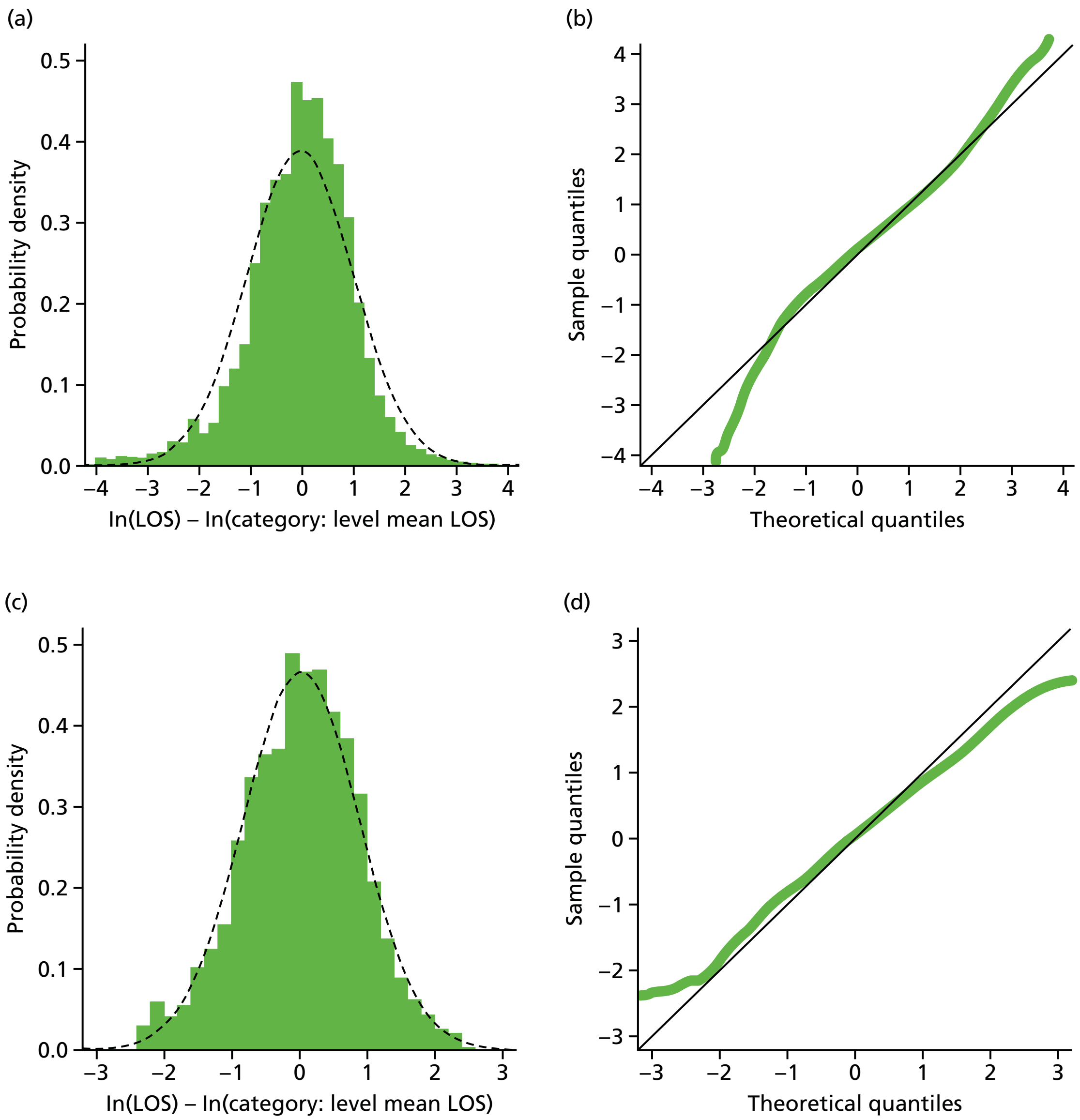


FIGURE 14 Comparison of actual data with an assumed log-normal distribution. IQR, interquartile range. (a) Density with no outlier removal (all data); (b) quantile-quantile plot with no outlier removal (all data: distribution density and distribution assuming log-normal distribution); (c) density with outlier removal [data with outlier detection (3.3% of all data removed by excluding points further than 1.5 × IQR from IQR): distribution density and distribution assuming log-normal distribution]; and (d) quantile-quantile plot with outlier removal [data with outlier detection (3.3% of all data removed by excluding points further than 1.5 × IQR from IQR)]. Each LOS is compared with the mean LOS for category of infant and level.

The log-normal distribution fitted the large majority (> 95%) of the data well. The actual data have a slight left-tail compared with the assumed distribution, showing a proportion of patients who have a significantly lower LOS in any particular level than the log-normal distribution would predict; however, this population is very small: 2.3% of episodes are lower than 1.5 × interquartile range (IQR) below the IQR. This cut-off point is used when actual LOS is < 9% of the category-level mean, or a log (ln) difference of < –2.4. Another 1% of all episodes have a LOS of > 1.5 × IQR above the IQR. Resulting lengths of stay used are shown in *Table 10*.

Length of stay in the model (when a level is used) uses a log-normal distribution as given in *Table 11*.

TABLE 11 Log (ln) LOS in a level

Parameter	Infant category	Log (ln) LOS (days)				
		Surgical	Level 1	Level 2	Level 3	Level 4
Mean	1	N/A	1.952	3.648	2.723	0.928
	2	N/A	2.652	3.575	3.195	0.766
	3	N/A	2.156	2.475	3.427	0.704
	4	N/A	1.208	1.376	3.101	0.657
	5	N/A	0.717	0.791	2.008	0.507
	6	N/A	0.697	0.397	0.768	0.104
	7	0.970	0.970	2.394	2.093	0.403
Standard deviation	1	N/A	2.057	1.150	1.225	1.020
	2	N/A	1.358	0.752	0.871	0.706
	3	N/A	0.894	1.038	0.536	0.685
	4	N/A	0.928	0.946	0.463	0.653
	5	N/A	0.886	0.833	0.936	0.648
	6	N/A	0.975	0.900	1.051	0.696
	7	1.491	1.491	1.362	1.251	0.776

N/A, not applicable.

Accuracy of model

Precision of model

The model had a warm-up period of 1 year, followed by 10 years of run-time. Variation between individual years in the base-case model (current configuration without resource constraints) were as follows:

- average distance from home – 0.6% coefficient of variation (CV)
- average number of infants in surgical phase intensive care – 6.4% CV
- average number of infants in Level 1 care – 3.0% CV
- average number of infants in Level 2 care – 1.1% CV
- average number of infants in Level 3 care – 0.6% CV
- average number of infants in Level 4 care – 1.1% CV
- average total number of infants in any neonatal care – 1.3% CV
- average nurse workload – 1.1% CV.

All 95% confidence limits of the mean estimated values above were less than ± 5% from the mean value. All results are presented as the mean across the 10 years.

Comparison between model and actual data

The average number of infants present and the average workload were compared between the model and the actual average admissions in the NDAU data. For this validation, the model was run with the assumption that all infants attend their closest appropriate unit.

The difference between predicted and actual admissions depended on the closeness of a unit to its nearest neighbouring unit (*Figure 15*). For units that were ≥ 15 minutes away from their nearest neighbouring unit, the accuracy of prediction of the number of infants present was typically ± 20–30%, or ± 2–3 infants, and prediction of nurse workload was typically ± 15–20% or ± 1 nurse equivalent workload.

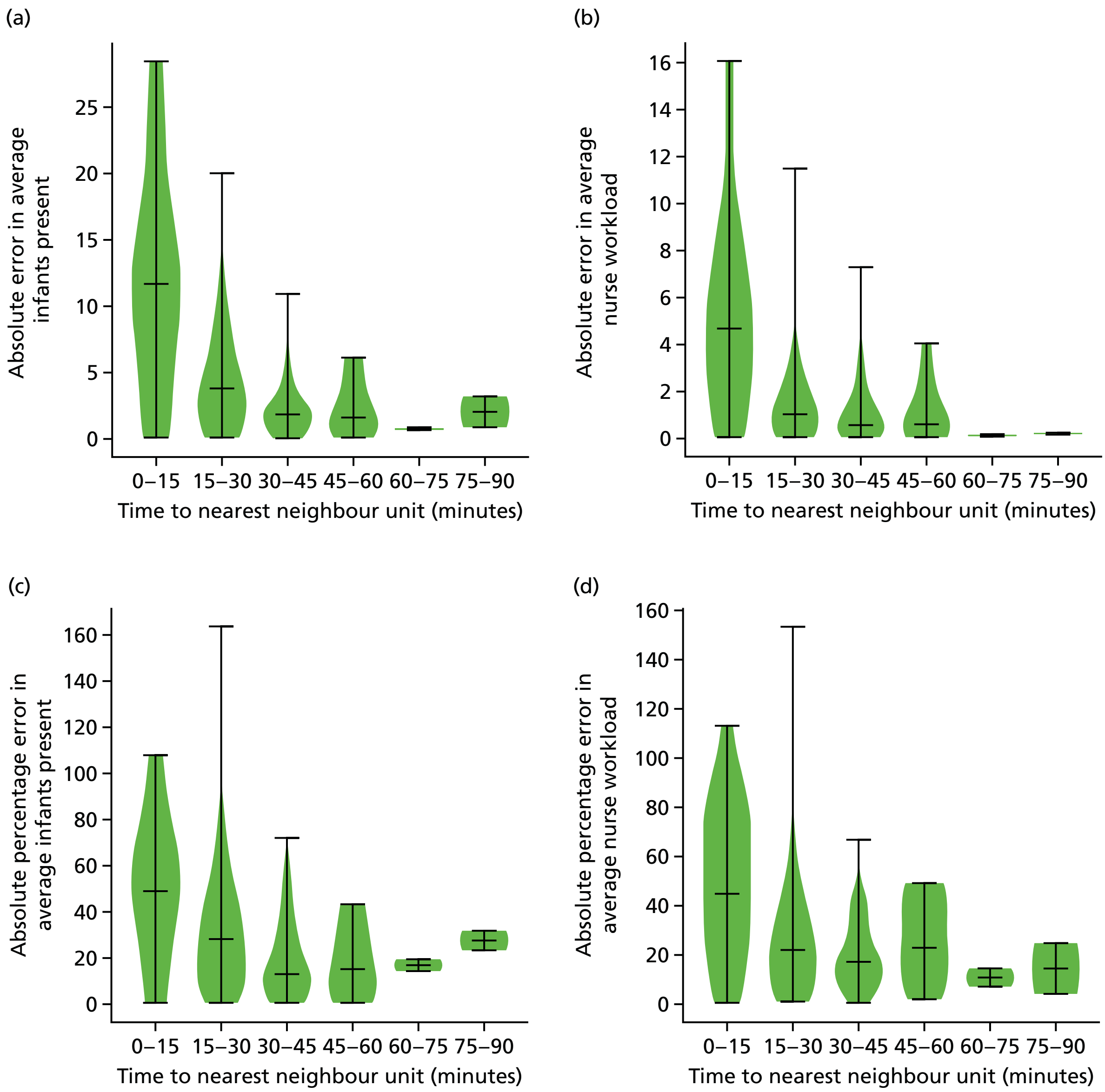


FIGURE 15 Violin plots showing accuracy of predicting neonatal unit occupancy and workload by proximity of a unit to its nearest neighbouring neonatal unit. Units are binned by proximity to the nearest neonatal unit: 0–15 minutes (18 units), 15–30 units (77 units), 30–45 minutes (35 units), 45–60 minutes (10 units), 60–75 minutes (2 units), and 75–90 minutes (2 units). Bars show the range of error in terms of absolute difference between model and actual occupancies and workload, with the middle cross-bar showing median error. The shaded regions shows distribution of error. The charts show error in occupancy [(a) and (c)] and nurse workload [(b) and (d)] expressed either as differences in numerical occupancy and workload [(a) and (b)] or percentage error from the actual occupancy and workload [(c) and (d)].

Median travel times from home to place of care compared well between the model and actual data. The median time from home for Level 1 care was 24 minutes in the model (compared with 21 minutes actual), Level 2 care was 14 minutes (compared with 16 minutes actual) and Level 3 care was 12 minutes (compared with 13 minutes actual).

Providing further credence to the principle of this geographic modelling, that infants will usually attend their closest neonatal unit (or one close to it), is that 95.3% of infants who only required local Level 3 care (special care) were cared for in either their closest unit or a unit no more than 15 minutes further than their closest neonatal unit.

Effect of altering capacity in the model

The model allows units to close to new admissions once a given nurse workload is reached (new admissions cannot take the unit above a defined capacity; so it is possible that an infant requiring Level 3 care may be admitted when an infant requiring Level 1 care has just been rejected). In the case of a unit not allowing an admission, the model searches for the closest unit with available space (defined by nurse workload) and that is the appropriate level.

Adverse effects on distance from home and the number of transfers are seen when average capacity utilisation increases to > 60% (*Figure 16*), with a doubling of the number of infants > 30, 45 or 60 minutes from home at approximately 75% of capacity utilisation.

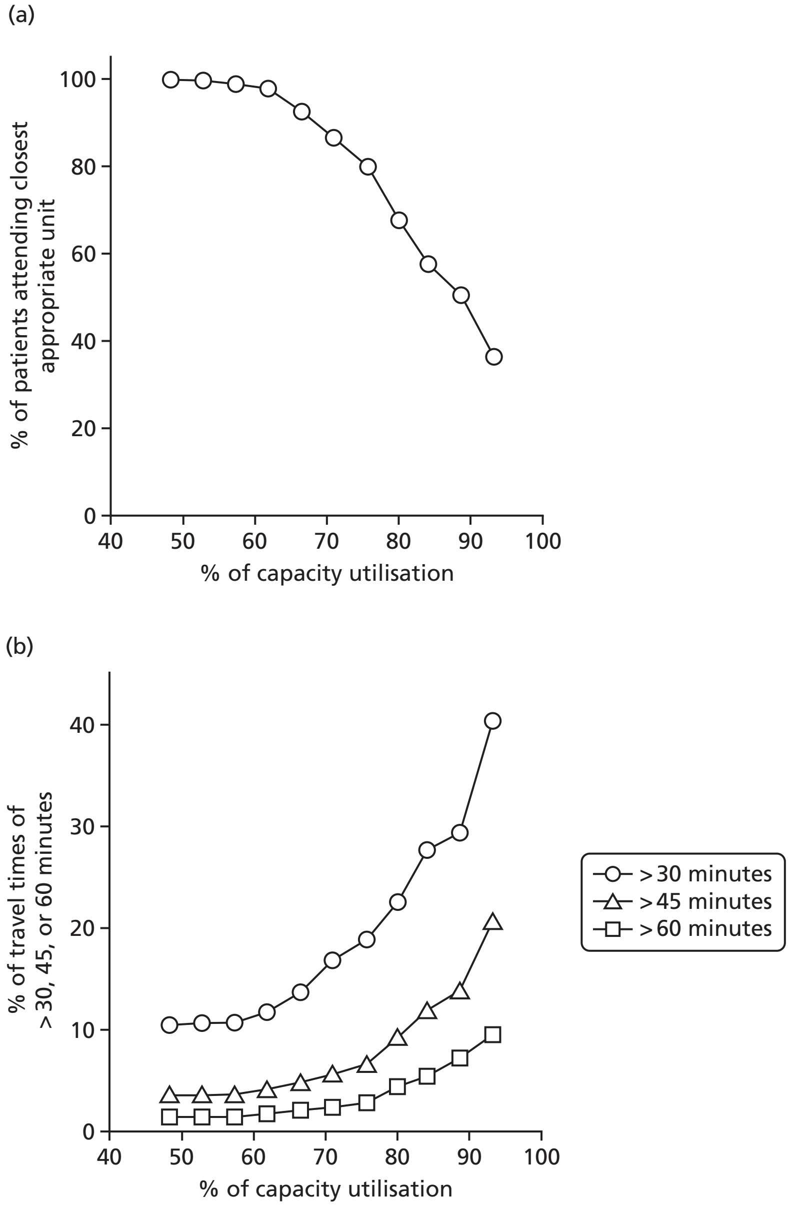


FIGURE 16 Effect of altering capacity on distance parents are from the place of care, and on the number of neonatal transfers. Capacity utilisation of 100% means that the unit is closed to further admissions. (a) The percentage of infants attending the closest appropriate unit; (b) the percentage of infants further than 30, 45 or 60 minutes from mother's home location; (c) the average travel time from mothers home location to place of care; and (d) the number of neonatal transfers (between neonatal units) per year. (*continued*)

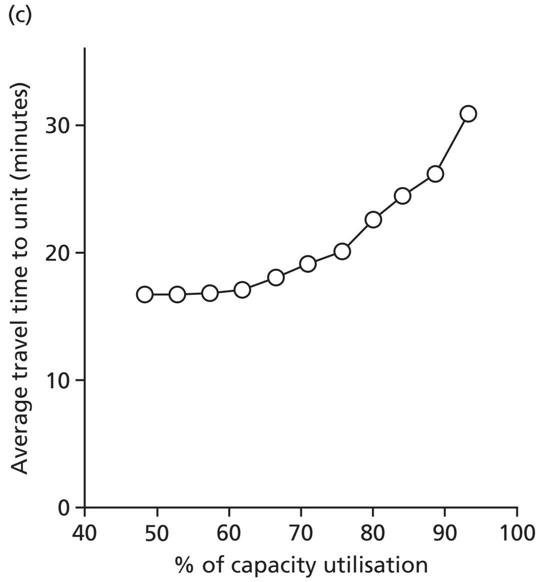


FIGURE 16 Effect of altering capacity on distance parents are from the place of care, and on the number of neonatal transfers. Capacity utilisation of 100% means that the unit is closed to further admissions. (a) The percentage of infants attending the closest appropriate unit; (b) the percentage of infants further than 30, 45 or 60 minutes from mother's home location; (c) the average travel time from mothers home location to place of care; and (d) the number of neonatal transfers (between neonatal units) per year.

Effect of unit size on variation in workload

The relative variation in workload depends on the size of the unit. As units increase in size, the relative variation in workload (ratio of peak-to-trough workload) reduces (*Figure 17*). Higher-volume units are therefore more robust against natural variation in workload.

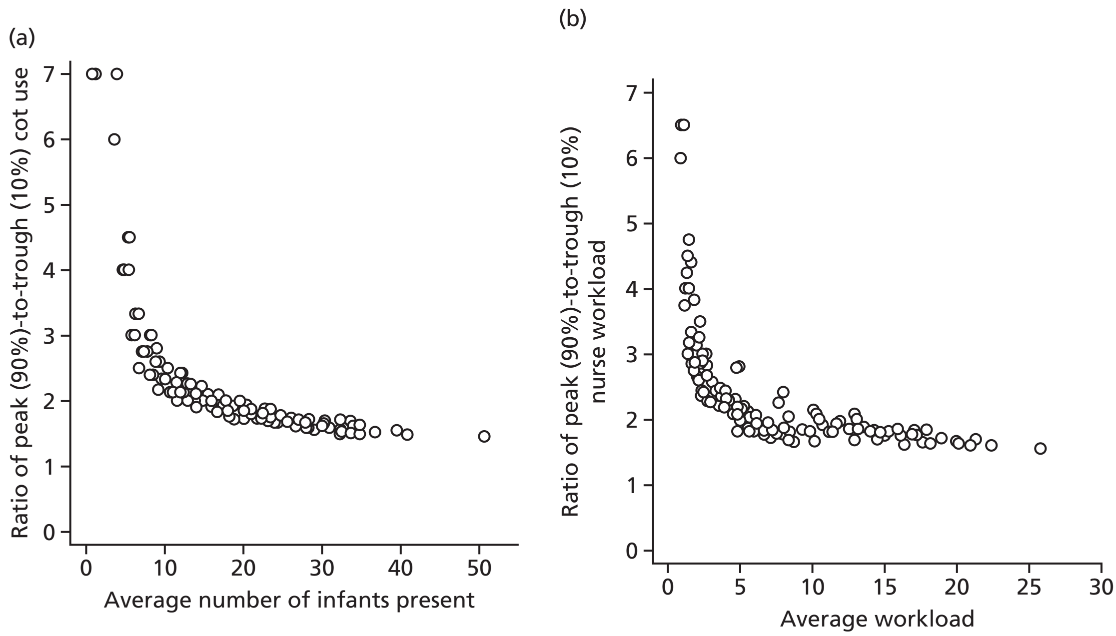


FIGURE 17 Variation, by hospital, (a) in cot use and (b) nurse workload by average cot use or workload. Variation was measured by calculating the ratio of common peak cot or nurse workload (90th percentile) to common trough cot or nurse workload (10th percentile). In two cases where the 10th percentile number of cots used or nurse workload was zero (giving an infinite workload ratio), the ratio is given a value of the maximum observed elsewhere.

Effect of removing network boundaries

In the base-case model, it was assumed that an infant would always go to their closest unit in their own network if capacity existed in an appropriate-level unit. The model was run without boundaries, such that an infant would always attend their closest appropriate unit that had capacity. Removal of network boundaries led to an improvement in infants cared for within 30, 45 and 60 minutes of their home location (*Table 12*). The difference was relatively small in absolute terms (typically an improvement of 0.5% to 1.0% of all patients in each travel band), but it is larger when expressed in relative terms.

Alternative scenarios

Two alternative scenarios, identified by the genetic algorithm (see *Chapter 6, Example of an alternative neonatal intensive care unit configuration* and *Example of a centralised neonatal intensive care unit configuration*), were tested in the simulation model. In one case, a configuration was picked that minimises travel distances while ensuring that all NICUs receive ≥ 100 VLBW infants per year. In the second case, we have chosen one example of significant centralisation (picking optimal locations for 30 NICUs, 30 LNUs and 30 SCUs). The scenarios were run either with (1) no resource constraints or (2) resources set so that units ran at an average of 80% of maximum capacity (*Table 13*). These scenarios were run without the specialist surgical LOS for those infants requiring surgery (although the rest of their stay that could be in non-surgical units was included). The surgery-specific workload, not included in this model, amounts to ≈24 on-duty nurse workload or approximately 2% of the total nurse workload in the model.

As expected from the genetic algorithm, the alternative scenario based on minimising travel distances while having all NICUs with ≥ 100 VLBW infants per year also reduced average travel times and increased the proportion of infants within 30, 45 and 60 minutes of the unit. It is worth noting that the aim of this scenario was primarily for all NICUs to be large enough to have ≥ 100 VLBW infants per year, so it is encouraging that travel distances may also be reduced simultaneously. This scenario also reduced the number of transfers and the transfer distance by ≈9%. The centralised scenario led to a significant increase in travel distances, but was able to reduce, by ≈9%, the number of nurses required to meet BAPM standards 90% of the time.

When capacity limits were placed in the system, such that units worked on average at ≈80% of maximum capacity (with capacity limited by nurses present), the performance of the system, viewed from the desire to have infants cared for in the closest appropriate unit, degraded. When running at an average of 80% of capacity utilisation, one-third of infant days were not in the closest appropriate unit in the two non-centralised scenarios. With the centralised scenario, which is more robust to variation in workload, 17% of patient care

TABLE 12 Effect of removal of network boundaries on travel times to place of care

Travel time to place of care	Existing network boundaries	No network boundaries	Relative increase in proportion of infants in band by using network boundaries
> 30 minutes (%)	10.5	9.8	6
> 45 minutes (%)	3.6	2.8	28
> 60 minutes (%)	1.5	1.1	33
Average (minutes)	16.7	16.3	N/A

N/A, not applicable.
Note
Data show the % of travel times from home in excess of 30, 45 and 60 minutes (from a semicontinuous audit of travel times in the model).

TABLE 13 Simulation of alternative configuration scenarios without resource constraints and with all units set to run at an average of 80% of absolute capacity

Model outputs	Current configuration (45 NICUs, 78 LNUs and 38 SCUs)	Alternative configuration[a] (48 NICUs, 78 LNUs and 35 SCUs)	Centralised configuration (30 NICUs, 30 LNUs and 30 SCUs)
No capacity constraints			
Mean daily travel time from home (minutes)	16	16	20
Infant days in planned place of care (%)	100	100	100
Patient days > 30 minutes from home (%)	9.4	8.2	14.6
Patient days > 45 minutes from home (%)	2.6	2.0	3.7
Patient days > 60 minutes from home (%)	0.9	0.8	1.3
Transfers per year (*n*)	17,213	16,008	17,347
Transfers per infant (*n*)	0.20	0.19	0.20
Transfer distance per year (miles, single direction)	360,986	314,151	396,723
Transfer time per year (hours, single direction)	10,188	9036	10,895
Average nurse workload present (*n*)	1062	1062	1062
On-duty nurses required to meet BAPM standards 90% of time (*n*)	1490	1512	1366
Average nurse utilisation to meet BAPM standards 90% of time (%)	71	70	78
Units planned to run at ≈80–85% of maximum capacity[b]			
Mean daily travel time from home (minutes)	22	21	23
Infant days in planned place of care (%)	67	67	83
Patient days > 30 minutes from home (%)	20.1	19.5	23.3
Patient days > 45 minutes from home (%)	8.0	7.1	8.8
Patient days > 60 minutes from home (%)	3.3	2.8	3.1
Transfers per year (*n*)	45,274	44,061	31,257
Transfers per infant (*n*)	0.51	0.48	0.34
Transfer distance per year (miles, singe direction)	966,975	898,714	846,293
Transfer time per year (hours, single direction)	26,767	25,418	21,815

TABLE 13 Simulation of alternative configuration scenarios without resource constraints and with all units set to run at an average of 80% of absolute capacity (*continued*)

Model outputs	Current configuration (45 NICUs, 78 LNUs and 38 SCUs)	Alternative configuration[a] (48 NICUs, 78 LNUs and 35 SCUs)	Centralised configuration (30 NICUs, 30 LNUs and 30 SCUs)
On-duty nurses[c] (*n*)	1333	1338	1296
Average nurse utilisation (%)	80	79	82

a Alternative configuration designed to minimise travel distances while having all NICUs receiving ≥ 100 VLBW infants per year.
b On-duty nurse recourses for each unit set at the next whole number up from a theoretical 85% capacity utilisation (e.g. if running at 85%, capacity is calculated as requiring 5.4 nurses present at any time then unit resources are set to six nurses at any one time).
c The number of on-duty nurses is given here assuming that capacity is capped at BAPM recommendations.[2] If there is allowed working beyond BAPM workload recommendations, then nurse numbers will scale down proportionally.

days were not in the closest appropriate unit. The increased robustness of the centralised scenario led to a closing of the gap in distances from home between that scenario and the two non-centralised scenarios; indeed, the centralised scenario had the fewest care days that were > 1 hour from the patient's home. The number of transfers was also lowest in the centralised scenario (it had been highest when resources were not limited at any unit).

The number of on-duty nurses in the centralised scenario is given assuming that capacity is capped at BAPM recommendations.[2] If there is allowed working beyond BAPM workload recommendations, then nurse numbers will scale down proportionally (e.g. if maximum unit capacity occurs when total workload is 30% above BAPM recommendations[2] then the number of nurses required would be divided by 1.3).

Simulation modelling discussion

There has been a variety of modelling and simulation work on neonatal care systems. This has included mathematical queuing models,[63–67] a simulation model[68] and a comparison of mathematical and system dynamics models.[69] We have also previously reported on a simulation model of a regional neonatal network.[33]

The simulation model presented here is the first one that models a national networked neonatal care system in which infants may move to different hospitals either for planned care or because of resource constraints in the most local appropriate unit.

The model contains some simplifications compared with the real world, for example:

- It is assumed that an infant's condition moves from worse to better; more-intensive levels of care take place first in the model, with no backwards movement from lower to high intensive care.
- The model assumes that the place of birth is in the closest hospital with any type of neonatal unit, but if higher levels of care are needed immediately then the infant's first place of care (following immediate transfer) will be in the higher-level unit.
- The model assumes that location of care is in the closest appropriate unit with capacity.
- The model assumes that choices on location are made based on estimated road travel time.

The model was run for a period of 10 years, after a warm-up period of 1 year. When model results were divided by year, it was found that the mean results had an expected precision of within ± 5% of the mean. There is therefore little error due to stochastic variation in the model runs. Median travel times to place of care were within ± 2 minutes (or within ± 10%) when comparing the model to the real world.

When hospital workloads were compared with modelled workloads (assuming that infants can always be cared for in their closest appropriate unit), the accuracy of the model depended on proximity between units. This suggests that modelling assumptions of an infant attending their closest appropriate unit are valid when there is reasonable travel time between units, but is less valid when units are close together (and when travel time might be less important in unit destination). Inaccuracy of demand in units that are close together does not significantly affect prediction of travel time to place of care as that will be similar and unaffected between choice of unit.

The effect of changing resources/capacity had a profound impact on the incidence of care away from the planned place of care. In a 200-bed hospital model, it was suggested that working at an average of 85% of maximum capacity was sufficient to avoid problems of lack of capacity.[8] In the neonatal system, the model predicts that running at a planned average of 85% of maximum capacity would lead to almost half of the patients being cared for away from their closest appropriate unit. The neonatal system has two key differences compared with Bagust *et al.*'s[8] work on hospitals. First, units are much smaller, and this causes greater relative variation in workload, making the unit more likely to exceed capacity more often than a whole hospital. Second, in the neonatal unit, exceeding capacity in one unit affects other units adversely; the missed admission to the first unit must be placed in another unit, reducing the destination unit's capacity for their own local demand. As the system becomes busier, the number of displaced infants (those infants not able to access the closest appropriate unit because of capacity constraints) increases and these displaced infants contribute to the possibility of excessive demand in the units in which they are placed. It is not always easy in the real world to define absolute maximum capacity. When capacity is limited by on-duty nurse staffing, the maximum capacity depends on local decisions on when to close a unit to further admissions – we have previously observed units sometimes working with half the number of nurses recommended by BAPM.[33] Spare capacity may therefore be found by accepting infants when staffing levels are significantly above the level recommended by BAPM, but creating capacity in such a way carries a risk of worsened outcome.[9] The occurrence of having to work at particularly high peaks in workload will be reduced by having fewer higher-volume units (but at the cost of increased travel times).

The effect of unit size on variability of workload was apparent in the simulation results: between-day variation in workload was significantly higher for smaller units. Variation is described as the ratio of peak-to-trough workloads, with a peak defined as the type of workload that can occur in the busiest 10% of days, and a trough defined as the type of workload that can occur in the quietest 10% of days. A unit with an average workload of 2–3 nurse equivalents was found to have a fourfold ratio of peak-to-trough nurse workloads, whereas a unit with an average workload of 10 nurse equivalents will typically have a twofold ratio of peak-to-trough nurse workloads.

There is, therefore, a significant tension in neonatal care. Neonatal care is expensive and financial challenges will tend to apply pressure to increase utilisation of these expensive resources. This pressure unavoidably leads to more neonatal transfers and care further away from home locations. It is not possible to combine high utilisation of neonatal care with care consistently provided close to home. The levels of utilisation (< 65% of the absolute maximum capacity) required to minimise the need for unplanned transfers are likely to be unacceptably low considering the financial pressures existing in health care. If high utilisation (such as ≥ 80%) of resources is required, then the system needs to be adapted to be able to manage the increased number of transfers, and consideration needs to be given to the number of parents for whom daily commuting to/from the place of care may become impossible. The number of parents who live > 60 minutes (driving) away from the location of care could be taken as an indicator of the group who may require overnight accommodation in or close to the hospital (possibly funded by the hospital or by the parents themselves). When capacity is not constrained, centralisation within 90 units might be expected to increase demand for overnight accommodation by about 40%. However, in a capacity-constrained system in the model, there was not an increase in this group of parents; indeed, there was a slight reduction attributable to the centralised system's better resilience to peaks in workload.

Smaller units are therefore likely to more frequently breach BAPM guidelines[2] or capacity limits unless they are staffed with a higher proportional spare average capacity.

The simulation model was used to investigate whether or not boundaries, which can potentially cause a patient to travel further than their closest clinically appropriate unit, had a significant effect on travel times. When removing boundaries, there is a very small (< 1-minute) improvement in average travel time. The number of infants who are > 45 or 60 minutes away is reduced by about 30%, but this is against a backdrop of only a small proportion of infants being over these times with network boundaries in place. Therefore, the model suggests that use of network boundaries has only a minor negative effect on travel times.

Two alternative scenarios were tested in the simulation model. An alternative configuration with a similar number of each type of unit was tested. This configuration was chosen for its ability to have all NICUs receiving ≥ 100 VLBW infants per year (linked with improved clinical outcome). As with the genetic algorithm results, this model also had slightly improved travel times. The model also showed that this configuration required about 10% fewer transfers. Therefore, modelling suggests that there are better configurations of care: configurations that can (1) improve travel time, (2) ensure that all NICUs are large enough to receive ≥ 100 VLBW infants per year and (3) required fewer transfers. A second model was tested to look at the effect of significant centralisation of care: centralisation to 90 units from the current 161. The centralised model had the disadvantage of higher travel distances under conditions in which capacity is not strained. This model did, however, show two significant advantages. First, the number of nurses required to meet BAPM standards 90% of the time for local demand was reduced by about 10% compared with the current configuration (attributable to the smaller relative fluctuations in workload that occur in these higher-volume units). Second, this model, when run with constrained resources, had half the number of displaced infants as the current configuration, with about a one-third fewer transfers. Travel distances in a resource-constrained system became similar to the more localised care configuration.

The performance of the system, from the perspective of travel distances and transfers, was found to be highly dependent on capacity within the system in all the models examined.

Geographic modelling and simulation has shown that there are potentially improved national configurations of neonatal care, increasing both access and the development of NICU centres of excellence with ≥ 100 VLBW infants per year. A practical issue is, then, how much of the gap, between the current configuration and a theoretically ideal solution, can be closed.

Chapter 8 Health economics modelling

Reorganisation of health-care services requires an evaluation of the clinical benefit and costs to the health service and to the wider society. The aims of this chapter are to provide the building blocks for this evaluation by exploring the impact that service reconfiguration has on clinical outcomes (mortality), costs (neonatal bed-days, LOS and parent costs) and to undertake qualitative research on the factors that families and policy-makers would like to see taken into consideration in determining service reconfiguration.

Clinical outcome

Literature review

During the last 20 years, many models have been developed to estimate infant and neonatal mortality. The majority of these models explored the impact of infant characteristics on mortality and estimated mortality for either the VLBW infants, weighing 800–1500 g, or very preterm infants, with a gestational age of 22–32 weeks. Most studies used a logistic regression approach with covariates that have been found to be strong predictors for neonatal mortality (i.e. of gestational age, small for gestational age, sex, birthweight and birthweight *z*-score).[70–73] A few studies considered sociodemographic and socioeconomic variables including race and ethnicity,[18,20,74–76] education, insurance status, percentage of inhabitants living below the poverty line[18,20,77] and the lowest decile of the IMD score.[19]

A smaller number of studies have investigated the impact of service configuration in terms of (1) working patterns and staffing and (2) organisational level. One of the first studies that looked at the impact of workload and staffing was by the Tucker and UK Neonatal Staffing Study Group,[78] which explored the impact of the availability of consultants (high availability defined as ≥ 2 consultants) and nurses relative to the BAPM nurse-to-cot ratio guidelines (high availability is defined as a nurse-to-infant ratio of ≥ 0.84). The UK Neonatal Staffing Study Group[78] used three workload measures, including occupancy, which measured the maximum number of infants present in a unit over their study period. The authors reported that for every 10% increase in percentage of maximum occupancy at admission, the odds of mortality increased by 1.09 (95% CI 1.01 to 1.18). Rautava *et al.*[79] explored the impact of working patterns on mortality and found that the risk of mortality for very preterm infants increased when the infant was born in non-office hours, and that mortality rates could be consequently improved by an increase of resources. Finally, Watson *et al.*[14] estimated the effect of the 1 : 1 nurse-to-patient ratio for intensive care neonates and found that a 1 : 1 nurse-to-patient ratio reduces infant mortality. A further set of studies explored the degree to which mortality varied depending on the care levels by which services were organised (e.g. NICUs, LNUs and SCUs in the UK). For example, Cifuentes *et al.*[20] estimated the mortality of low-birthweight infants for different levels of care in the California area and found that the level of care in NICUs can influence the probability of survival. The 2010 systematic review by Lasswell *et al.*[80] explored the association between the designation level of hospital and VLBW infant mortality based on studies that evaluated the regionalisation of perinatal services for very preterm or VLBW infants. Most studies that looked at the impact of organisation in terms of staffing or hospital level used logistic regression, with the exception of Watson *et al.*,[19] who used an instrumental variable approach. Care is needed in interpreting estimation of the logistic model when there is a small number of events, as is the case in neonatal mortality, and so these estimates may be affected by small-sample bias.

Other studies have aimed to look at the impact of organisational factors, such as the number or volume of infants treated in neonatal units, based on the idea that staff can become more experienced and skilful as they treat a larger number of different and complex infants. A high-volume unit has been defined as one that treats at least a fixed number of VLBW or very preterm infants per year[76,78] or as one that is in the top quartile of all neonatal units.[19] Rogowski *et al.*[76] explored the impact of hospital-level determinants of mortality among VLBW infants and showed that the number of VLBW infants admitted to a neonatal unit reduced infant mortality. More recently, studies have aimed to look at the causal relationship between infants born in high-volume units and infant mortality using instumental-variable (IV) approach methods.[14,19,77] The effect of designation and volume of neonatal care for preterm birth reported in Watson *et al.*[19] had a significant effect, especially on neonatal mortality and in-hospital morbidities (bronchopulmonary dysplasia, necrotising enterocolitis and retinopathy of prematurity). For infants with < 33 weeks of gestational age, the risk of death was reduced by 2.6 percentage points if admitted to a high-volume unit, and this effect was higher if infants had a gestational age of < 27 weeks. Finally, some studies explored the impact of hospital volume within different levels of care. For instance, Phibbs *et al.*[18] examined the impact of hospital volume among different levels of neonatal care units and found that volume and level had a significant impact on risk of infant mortality, showing that the delivery of VLBW infants in NICUs with a high volume can reduce neonatal mortality.

Table 14 summarises the neonatal mortality models that explored the impact of volume on mortality.

Thus, the few studies that have attempted to estimate causal effects suggest that there is a positive effect of birth in higher-volume hospitals, whereas there is no evidence that birth in a hospital with a NICU has any effect on mortality. However, previous studies have only compared NICUs against all other hospital designation categories combined without distinguishing between SCU and LNU hospitals. Furthermore, there is no study in England that has evaluated the causal impact on LOS and reimbursement costs. Our aim was to estimate the impact of volume and designation level on mortality and costs between NICUs, SNUs and LNUs, and separately between high-volume units and other hospitals of birth.

Data

Data relative to neonatal care were collected in 2014/15 from units in England as part of the BadgerNet data set. The distance in time and miles was evaluated using LSOAs of the mother and the postcode of the hospital. Mortality was defined as mortality during the in-hospital period from the admission to the discharge.

Mortality was registered between 2014 and 2015 for a total of 2010 infants, out of a total number of 165,450 admissions to neonatal units. Out of all registered deaths, 52% were for infants born with a gestational age of < 28 weeks. By adding infants born between 29 and 32 weeks of gestational age, 65% of deaths are covered; including infants born between 33 and 36 weeks, up to 83% of deaths are accounted for. *Figure 18* illustrates the rate of death and survival per gestational age of infants admitted to neonatal units in our study.

Method

High-risk infants tend to be treated in high-volume units, so in order to estimate the effect of volume on mortality, it is important to measure this effect in a representative sample of infants, including both high- and low-risk infants. More recent studies have aimed to estimate the causal effect of volume using an IV approach to control for confounding. The proximity to care can be used as an instrument that determines the chances of receiving care (e.g. birth in a hospital with a NICU) and should be independent of infant mortality. Proximity to care can be used to control confounding by estimating the difference in mortality between those who live close to NICUs and those who live far from NICUs, and both groups should be made up of a comparable mixture of high- and low-risk infants. The strength of an instrument can be tested by looking at the relationship between the instrument (i.e. travel time) and attendance at high-volume NICUs. In our case, a strong instrument would imply that those who live nearest to high-volume units are more likely to attend at those hospitals (i.e. the t-statistic on travel time, or the F-statistic if there is more than one instrument, is > 10).

Watson *et al.*[19] used an IV approach based on travel distance alongside eight other instruments [i.e. surgical facilities (1 = yes, 0 = no), high volume (1 = yes, 0 = no), Level 3 (1 = yes, 0 = no), Level 2 (1 = yes, 0 = no), distance × surgical facility, distance × high volume, distance × Level 3 and distance × Level 2]. Level 3 represents NICUs and Level 2 represents LNUs.

TABLE 14 Brief description of effect of volume on neonatal outcomes reported in the literature

Study, author, year (country)	Volume	Mortality	Effect, OR (95% CI)	Description
Tucker and UK Neonatal Staffing Study Group,[78] 2002 (UK)	• High (> 57 VLBW infants) • Medium (35–57 VLBW infants) • Low (< 35 VLBW infants)	There are no differences in the odds of mortality HV units treat sicker infants than MV and LV units do	• HV 1.0 • MV 1.12 (0.76 to 1.64) • LV 0.97 (0.70 to 1.34)	Generalised estimating equation model (logistic)
Watson *et al.*,[19] 2014 (UK)	HV for units with ≥ 100 VLBW infants per year Tertiary units are NICUs	TUs are NICUs Consistent reduction in OR of mortality for very preterm infants admitted to HV neonatal units	• TU 0.77 (0.59 to 1.00) for < 32 weeks' GA • TU 0.65 (0.46 to 0.91) for < 26 weeks' GA • TU 0.92 (0.69 to 1.22) for < 27–31 weeks' GA (reported in paper) • HV 0.73 (0.56 to 0.95) for < 32 weeks' GA • HV 0.62 (0.44 to 087) for < 26 weeks' GA • HV 0.86 (0.65 to 1.14) for < 27–31 weeks' GA (reported in paper)	IVs
Gale *et al.*,[81] 2012 (UK)	HV: unit with ≥ 2000 neonatal intensive care-days annually	Centralisation of neonatal intensive care within a smaller number of neonatal units providing both a high level of intensive care and HV of activity is associated with reduced mortality	• (HV is the reference) • LV (≤ 2000 IC days) 3.27 (2.92 to 3.66) • HV (> 2000 IC days) OR 1.0	Logistic regression analysis
Cifuentes *et al.*,[20] 2002 (USA)	Patient of VLBW infants: • HV PV ≥ 15 • LV PV < 15 Four levels of NICU: 1. no 2. intermediate 3. community 4. regional	In higher levels of NICUs when volume is considered, there is a marked reduction in average mortality risk	• (Regional NICU is the reference) • Community NICU BW < 2000 g, PV ≥ 15 1.11 (0.87 to 1.43) • Community NICU BW < 2000 g, PV < 15 1.42 (1.14 to 1.76) • Community NICU BW < 1500 g, PV ≥ 15 1.05 (0.77 to 1.44) • Community NICU BW < 1500 g, PV < 15 1.51 (1.14 to 2.00) • Community NICU BW < 1250 g, PV ≥ 15 1.08 (0.76 to 1.54) • Community NICU BW < 1250 g, PV < 15 1.48 (1.07 to 2.05)	Logistic regression analysis

continued

TABLE 14 Brief description of effect of volume on neonatal outcomes reported in the literature (*continued*)

Study, author, year (country)	Volume	Mortality	Effect, OR (95% CI)	Description
Rogowski *et al.*,[76] 2004 (USA)	Patient volume of VLBW infants: 50 Three levels of NICUs: 1. restriction on ventilation, minor surgery only 2. major surgery 3. cardiac surgery	Volume and NICU level explain very little of the variation across hospital in mortality among VLBW infants	• HV (≥ 50) 0.989 (0.983 to 0.994) • LV (< 50) 1.001 (1.000 to 1.002)	Random-effect logistic regression
Phibbs *et al.*,[18] 2007 (USA)	Five levels: • 1 – No NICU • 2 – NICU provides care for mildly ill infants but not mechanical ventilation • 3A – NICU provides mechanical ventilation with restrictions • 3B – NICU provides mechanical ventilation without restrictions but does not provide major surgery • 3C – NICU provides major surgery but not open-heart surgery and ECMO • 3D – NICU provides cardiac surgery requiring cardiopulmonary bypass or ECMO PV (10, 25, 50, 100)	The effects of the volume of VLBW infants varies depending on the NICU level. Mortality decreases as patient volume increases within each level of care and with higher levels of care within each volume group	• Level 1 PV (< 11) 2.72 (2.37 to 3.13) • Level 1 PV (> 10) 2.39 (1.91 to 3.00) • Level 2 PV (< 11) 2.53 (2.02 to 3.18) • Level 2 PV (11–25) 1.88 (1.56 to 2.26) • Level 2 PV (25) 1.22 (0.98 to 1.52) • Level 3A PV (< 26) 1.69 (1.28 to 2.24) • Level 3A PV (26–50) 1.78 (1.35 to 2.34) • Level 3A PV (> 50) 1.08 (0.96 to 1.21) • Level 3B, 3C PV (< 26) 1.51 (1.17 to 1.95) • Level 3B, 3C PV (≥ 25) 1.30 (1.12 to 1.50) • Level 3B, 3C, 3D PV (51–100) 1.19 (1.04 to 1.37) • Level 3B, 3C, 3D PV (> 100) OR 1.00	Multiple logistic regressions

BW, birthweight; ECMO, extracorporeal membrane oxygenation; GA, gestational age; HV, high volume; IC, intensive care; LV, low volume; MV, medium volume; PV, patient volume; TU, tertiary unit.

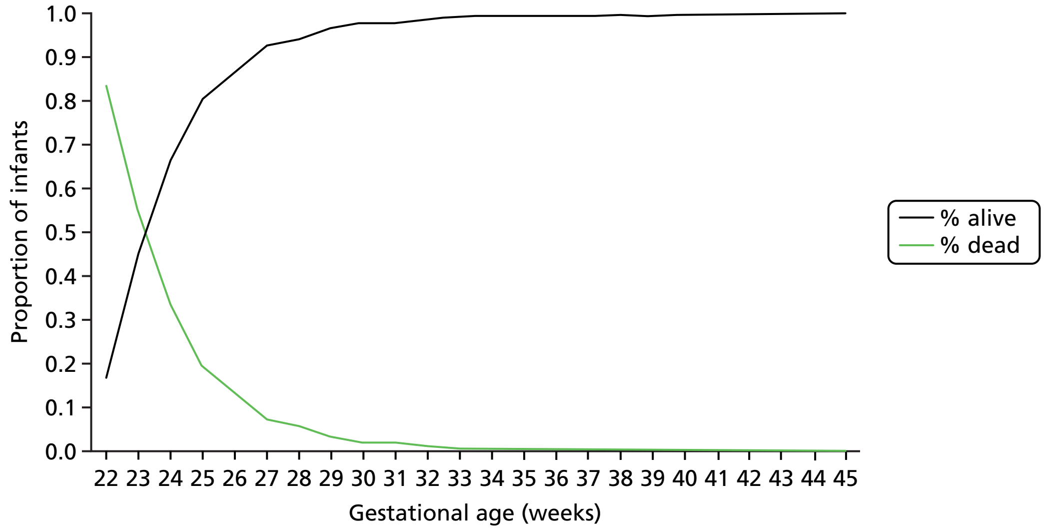


FIGURE 18 Mortality and survival rates of infants by gestational age.

The main analyses will use only one instrument (travel time or travel distance) to facilitate interpretation and to eliminate the need to identify and remove weak instruments when multiple instruments are used. The secondary analysis will use multiple instruments. We control for the following covariates in the model: age and age squared at birth, sex, deprivation of residence (quintiles of multiple deprivation), mode of delivery (emergency caesarean without labour, emergency caesarean with labour, vaginal non-spontaneous, elective section, unknown or vaginal spontaneous) and fetus number. A similar approach is used to estimate the impact of high volume on total LOS (sometimes referred as the super stay) and the associated reimbursement costs by level of care (BAPM) actually received, for infants referred to high-volume units, where length of hospital stay is defined as the number of days from admission to hospital discharge or death, whichever took place first. The LOS results are shown in the evaluation section of this report (see *Chapter 9*).

Two sets of neonatal mortality models are estimated: semiparametric and parametric models. The semiparametric model is a structural mean model (SMM)[82] and serves to estimate the treatment effect on the treated, thus allowing for the possibility of different treatment effects between the treated (NICU-born) and untreated (non-NICU-born) infants. The parametric model instead adopts a bivariate (probit) distribution for mortality outcomes and the exposure status (birth in a hospital with a NICU vs. birth in a hospital without a NICU) and allows us to estimate the average treatment effect (i.e. the effect in the whole infant population, at the cost of imposing the assumption of homogeneity of treatment effect and normal distribution of unobservable confounding factors). We present both sets of results, but in the main discussion we focus on the semiparametric results, given that these results are based on less restrictive assumptions and so are more robust estimates of the effect. We have instrumented both the semiparametric and parametric models using an IV approach and, for comparison, run a linear probability model (LPM) that has no instrument [i.e. ordinary least squares (OLS)].

Two different units were considered: high-volume and tertiary units (i.e. NICUs). High-volume units are characterised by a minimum number of 100 admissions per year of infants with a birthweight of < 1500 g. Tertiary units are represented by NICUs that are the highest level of neonatal care, providing a service dedicated to babies needing respiratory support (ventilation) weighing < 1000 g, born at < 28 weeks' gestation or needing significant continuous positive airway pressure support.

Like Watson *et al.*,[19] we estimated infant mortality for infants born at a gestational age of < 32 weeks in a high-volume unit or hospital with a NICU. In addition, we conducted analysis to explore the effect of neonatal transfers to a NICU hospital from a lower-level hospital in infants born before 32 weeks' gestational age. We conducted a sensitivity analysis on the results for those born between 26^{+0} and 31^{+6} weeks of gestational age, to account for the possibility of bias due to the effect of imbalance in the distribution of extremely premature babies across treatment (hospital of birth) groups.

Secondary analyses of the relative effects of birth in a hospital with a NICU versus a SCU and a LNU were conducted using three available instruments of travel time to these three types of hospitals. These IV models were formulated as seemingly unrelated equations and estimated by simulated maximum likelihood.[83] In these analyses, neonatal mortality was analysed using multivariate probit distributions. Because we did not have complete information on the closest LNU and SCU units to some infants in the data set, we conducted these analyses excluding infants with incomplete data.

Results

We first checked if travel time was correlated with the exposure variable (i.e. birth in a hospital with a high-volume neonatal unit, and birth in a hospital with a NICU), thus supporting its use as an IV. Second, we looked at whether or not travel time is correlated with any other sample characteristics, such as birthweight and sex, to check if any possible effect of travel time on mortality operating through the exposure variable is confounded by other variables.

The descriptive statistics in *Table 15* summarise sample characteristics by tertiles of travel time for high-volume units and NICUs and shows that in most cases there are no systematic differences of sample characteristics across the travel time tertiles. In addition to the exposure variables (delivery at hospital with a NICU and delivery at hospital with a high-volume unit), systematic differences arise only for deprivation of residence and unknown delivery mode; this suggests the need to control for possible confounding by these variables in our analyses.

TABLE 15 Sample characteristics by travel time to high-volume units and NICUs

Outcomes and sample characteristics	Occurence of outcomes and sample characteristics by travel time to high-volume units and NICUs (%)		
Travel time to high-volume units	***Lower tertile (n = 4187)***	***Medium tertile (n = 4188)***	***High tertile (n = 4346)***
Died	7.45	8.62	9.19
Discharged to home	87.34	84.99	81.00
Discharged to ward	1.38	1.60	1.90
Last record: transferred to another hospital/unit	3.32	4.33	7.52
Unknown destination	0.52	0.45	0.39
Gestational age at birth (weeks)	28.40	28.49	28.43
Birthweight (kg)	1.19	1.21	1.20
≥ 2 fetuses	26.39	27.39	27.36
Female sex	46.62	45.22	46.38
Residence: most deprived quintile	41.01	27.96	29.73
Residence: 2nd most deprived quintile	26.41	22.13	20.69
Residence: 3–5 least deprived quintile	32.57	49.90	49.58
Caesarean delivery	49.15	50.93	50.57
Spontaneous vaginal	36.52	36.34	37.75
Unknown delivery mode	5.85	2.79	2.99
Delivery at high-volume unit	37.76	14.06	10.78

TABLE 15 Sample characteristics by travel time to high-volume units and NICUs (*continued*)

Outcomes and sample characteristics	Occurence of outcomes and sample characteristics by travel time to high-volume units and NICUs (%)		
Travel time to a hospital with a NICU	***Lower tertile (N = 4191)***	***Medium tertile (N = 4185)***	***High tertile (N = 4311)***
Died	8.26	8.89	8.14
Discharged to home	87.30	84.49	81.52
Discharged to ward	1.29	1.53	2.07
Last record: transferred to another hospital/unit	2.94	4.56	7.66
Unknown destination	0.21	0.55	0.60
Gestational age at birth (weeks)	28.41	28.43	28.47
Birthweight (kg)	1.19	1.20	1.21
≥ 2 fetuses	25.84	27.60	27.70
Female sex	46.36	45.16	46.69
Residence: most deprived quintile	47.67	29.49	21.76
Residence: 2nd most deprived quintile	23.00	24.35	21.87
Residence: 3–5 least deprived quintiles	29.33	46.16	56.36
Caesarean delivery	48.34	50.75	51.54
Spontaneous vaginal delivery	37.06	36.92	36.67
Unknown delivery mode	4.84	3.70	0.00
Delivery at hospital with NICU	81.53	42.39	26.70
Delivery at hospital with LNU	13.36	47.22	58.76
Delivery at hospital with SCU	5.11	10.39	14.54

Note
In the sample, lower and higher tertiles of travel time to high-volume units are 31 and 56 minutes, respectively; the lower and higher tertiles of travel time to NICUs are 17 and 31 minutes, respectively.

Table 16 shows the mortality model using LPM, semiparametric IVs and parametric IV bivariate probit model (marginal effect) for the high-volume units and hospitals with NICU. The controlled covariates used in these models are gestational age and age squared at birth, birthweight, sex, deprivation of residence, mode of delivery and fetus number.

The instrument strength reported for the IV model estimates, both in the linear SMM and bivariate probit models, shows that the instrument is strong (e.g. the *t*-score test statistic on travel time is 32 for high-volume units; rule of thumb is that a robust instrument has an *F*-test statistic of > 10). In addition, travel time affects the treatment variable (the probability of delivery at a hospital with a high-volume unit in one analysis and delivery at a hospital with a NICU in another) in the expected direction and is negative, implying that the longer travel times are associated with a lower likelihood of birth in hospitals with high-volume units and of birth in hospitals with NICUs.

The estimated causal effects of the IV models indicate that delivery in high-volume units reduces neonatal mortality by 1.2 percentage points in accordance with the bivariate probit model and by 5.0 percentage points with the linear SMM. In contrast, the LPM reports high-volume units having an increased risk of death, but this result is likely to be affected by confounding issues (i.e. high-volume units treat high-risk

TABLE 16 Effect of neonatal care at high-volume units and NICUs ($n = 12,687$)

Estimated effect/parameter/test statistic	LPM	IVs linear SMM	IV (marginal effect) bivariate probit model
Causal effect on mortality of neonatal care at hospitals with ≥ 100 babies weighing 1500 g per year			
Delivery at high-volume unit, absolute risk difference vs. non-high-volume unit (SE)	0.009 (0.006)	–0.050** (0.020)	–0.012*** (0.004)
Minimum travel time (minutes) to high-volume unit, coefficient (SE)	N/A	–0.003*** (0.000)	–0.018*** (0.001)
Instrument strength: *t*-/*z*-test statistic	N/A	32.2	32.0
Hausman test χ^2 statistic of H0: no endogeneity of treatment variable	N/A	N/A[a]	15.3***
Causal effect on mortality of neonatal care in hospitals with a NICU			
Birth at hospital with NICU	–0.009** (0.005)	–0.012 (0.012)	–0.006 (0.007)
Minimum travel time (minutes) to NICU	N/A	–0.010*** (0.000)	–0.031*** (0.001)
Instrument strength: *t*-/*z*-test statistic	N/A	39.8	41.8
Hausman test χ^2 statistic of H0: no endogeneity of treatment variable	N/A	N/A[a]	0.1

*$p < 0.10$; **$p < 0.05$; ***$p < 0.01$.
H0, null hypothesis; N/A, not applicable; SE, standard error.
a This model was estimated using the Generalised Method of Moments and so the Hausman test is not applicable.
Note
Controlled covariates: age and age squared at birth, birthweight, birthweight squared, sex, deprivation of residence, mode of delivery and fetus number.

infants and so are likely to have higher mortality), as indicated by the Hausman test statistic. Sensitivity analyses excluding infants born at < 26 weeks' gestational age were also conducted and they confirmed these findings (see *Appendix 2*).

Birth in a hospital with a NICU does not appear to result in any difference in terms of mortality relative to other hospitals, as summarised in *Table 13*. These results are based only on the use of one instrument: travel time to closest NICU hospital. We ran additional analysis to explore the effectiveness of NICUs for those infants born in a hospital with a NICU or transferred within the first 48-hour period. Of all neonatal transfers with a recorded transfer time ($n = 1519$), 65% took place within 48 hours of birth. Infants who were born in a hospital with a NICU or who were transferred to a hospital with a NICU had no detectable effect on mortality, using the IV approach based on the single instrument of travel time to closest NICU (or distance to closest NICU). The same results were obtained when the sample was limited to those infants born between 26^{+0} and 31^{+6} weeks' gestational age (see results in *Appendix 2*).

Secondary analysis of the relative effects of birth in a hospital with a NICU compared with a hospital with a SCU or a LNU were conducted using three available instruments of travel time to these three types of hospitals. In these additional analyses, we find that the NICU does appear to reduce mortality, compared with the other levels of care, by 2 percentage points, and so suggests that NICUs in themselves have some beneficial impact on mortality compared with other levels of care (see *Appendix 2*).

Discussion

We estimate infant mortality for infants born at a gestational age of < 32 weeks as a function of exposure to high-volume unit or hospital with a NICU at birth. We find that exposure to a high-volume unit at birth reduces mortality relative to other neonatal units. A very preterm infant born in a high-volume unit (≥ 100 babies weighing < 1500 g per year) has a 5-percentage-point lower risk of death in this unit than in other neonatal units when travel time is used as the instrument and mortality is estimated using a semiparametric method (linear SMM). This estimate drops to 1.2 percentage points when the same analysis is run using a parametric method (bivariate probit). There is a debate to be had about which result should be given greater credence. The semiparametric approach is based on less-restrictive assumptions and, therefore, is potentially more robust to violations of assumptions underpinning the analysis, so we have chosen to emphasis this result here; to allow comparisons with existing literature in this area, it is necessary to further discuss the parametric result. The parametric result is also the one that we use in the evaluation section of this report to calculate the incremental cost-effectiveness of high-volume units, to avoid potential problems with predicted values outside the 0–1 probability range.

Watson *et al.*[19] found that a preterm infant born in a high-volume unit (defined as those in the top quartile of all neonatal units) has a 2.6-percentage-point lower risk of death than in other neonatal units when travel distance is used as the instrument and mortality is estimated using a parametric method. This percentage point risk reduction is not immediately apparent from the paper, but can be calculated from the reported OR of 0.68 for in-hospital mortality reported in the paper (which approximates the risk ratio in cases like this when the deaths are rare), and the in-hospital mortality for high-volume units reported as 5.5 percentage points in the descriptive statistics [giving an estimated percentage point reduction of approximately = (5.5/0.68) – 5.5 = 2.6 percentage points]. The 2014 estimate of Watson *et al.*[19] is higher than the 1.2 percentage point reduction found by our parametric approach. An obvious explanation for the differences is the definitions used for high-volume units, but similar results were found when we defined high volume as those in the top quartile of all neonatal units. Another reason for the differences is the instruments used, given that Watson *et al.*[19] used travel distance and we explored the use of both travel distance and travel time. We report here only the results based on travel time as an instrument because there is strong support for travel time accurately representing access to health-care services. Sensitivity analysis excluding infants born at < 26 weeks of gestational age halves the mortality effect of birth in high-volume units compared with other units (2 vs. 5 percentage points in all the infants aged < 32 weeks' gestational age), but the estimates are imprecise.

A baby being born at < 32 weeks' gestational age in a hospital with a NICU does not appear to result in any difference in terms of the risk of death compared with other units. Similar results are also found by Watson *et al.*[19] We ran additional analysis to explore the effectiveness of NICUs for those infants born in a hospital with a NICU or transferred within the first 48-hour period. Birth at a hospital with a NICU or being transferred to a NICU within a 48-hour period had no detectable effect on mortality when using the IV approach based on the single instrument of travel time to closest NICU (or distance to closest NICU).

In our interviews, policy-makers raised queries about the robustness of the finding that NICUs did not affect the risk of death compared with other hospitals. To check the robustness of the result, we present additional analyses comparing the effectiveness of NICUs with other levels of care (i.e. NICU vs. LNU and NICU vs. SCU). All other results for the NICU were based on only one instrument: travel time to closest NICU. The advantage of considering other levels of care is that it opened up the approach to using three instruments: travel time to the closet NICU, LNU and SCU. A slight disadvantage of the approach is that the data set was less complete, because we did not have complete information on the closest LNU and SCU for some infants in the data set; therefore, we were restricted to performing these additional analyses on a smaller data set that excluded infants with incomplete data. In these additional analyses, we find that NICU does appear to reduce mortality by 2 percentage points, compared with the other levels of care.

A limitation of the data and analysis is that it is currently not possible to estimate the impact of mortality for those infants who are transferred into NICUs. The numbers of transfers are not insignificant; for example, for all infants with a gestational age of < 33 weeks at birth and who have ≥ 7 days of BAPM Level 1 (intensive) care, 61.2% are born in a hospital with a NICU, 18.9% are born in a hospital without a NICU and are transferred to a NICU and 19.8% are born in a hospital without a NICU and are not transferred to a NICU. We are therefore unable to separate out the benefits of antenatal care taking place prior to birth. It is clear that birth in a high-volume unit leads to improvements in mortality, and closing down low-volume units might lead to more infants being born in high-volume units, but the impact of changes in transfers is unclear.

Costs

In this part of the study we had planned to look at NHS neonatal costs in more detail by first gaining a better understanding of the national reference costs and how they inform HRGs. We had also planned to assess the components that make up the national reference costs by collecting data from the four main types of neonatal units that currently exist in the UK, and explore how these data are seen to vary by the number of infants. Finally, we had planned to estimate the costs of neonatal care for families, based on a survey of family costs by BLISS.

When looking at the national reference costs[84] and how they inform HRGs, it became clear that units were still paid in accordance with the HRG 2001 data set,[85] which did not accurately reflect resource usage by BAPM guidelines[2] (i.e. nurse-to-infant ratios). The reference cost submissions in July 2017[86] were the first to ask for units to submit data in accordance with the revised HRG reference cost guidance that took BAPM 2011 guidelines[2] into account. During the course of our study and interviews with unit staff, a lot of units were still trying to work out how best to apply the new guidance and so were reluctant to release cost data, making it hard to apportion costs to the different activities. To assess the impact on NHS costs, we decided to shift the focus from assessing the components that make up the reference costs to exploring the impact of high volume on the LOS of infants, to allow us to begin to explore the impacts of reorganisation. Further work on the cost components would be possible using the submissions under the new guidance that became available in January 2018, but which is beyond the scope of this current report.

Literature review

There are a number of papers that look at the impact of gestational age on hospital neonatal costs and families. Rogoswki *et al.*[87] explored the impact of gestational age on neonatal and perinatal cost in the American Vermont Oxford hospital network for NICUs. Costs were classified into accommodation costs and ancillary costs; ancillary costs were divided into five subcategories: respiratory therapy, laboratory, radiology, pharmacy and other ancillary. The authors show how costs vary within gestational age, birthweight, location of birth and discharge status. The category of infants who have the highest costs are those born between 24 and 26 weeks, with a birthweight of < 1000 g and born outside the hospital. The study also shows an inverse relationship between costs and gestational age; this result is also confirmed for neonatal and childhood costs for extreme preterm[88] and preterm births.[89] Further work by Petrou *et al.*[88] estimated costs for extremely preterm birth for families using evidence from a population study. Results show that extremely preterm births are associated with higher public sector costs and there is an inverse relationship between costs and gestational weeks. Several sociodemographic covariates were included in the model, but only long-term unemployment is associated with an increase in costs.

For service reorganisations, it is important to consider the impact on costs as services change and the effects of economies of scale and scope. One approach used to address this question is to look at the elements that make up the costs and analyse how they vary by case mix. The UK Neonatal Staffing Study Steering Group developed a cost function to evaluate the nature and the degree of economies of scale in the provision of care for NICUs.[90] The economic analysis shows that volume and case mix interact to determine the degree of economies of scale, even if the determinants of costs and efficiency in neonatal costs have a high complexity. The treatment of the sickest infants centralised at a regional level can take advantage of economies of scale. Another study by O'Neill *et al.*[91] investigated the relationship between activity (total days of care provided and total days of intensive care provided) and costs (clinical staffing, support services and overheads) using a multivariate regression model. They found an inverse relationship between average cost per day and scale of services provided, confirming the benefits of centralisation of intensive care in larger units. The authors[91] also show that the adoption of a different form of estimation (i.e. the log–log or double-log function) provided the best fit to the data.

Another approach is to simply look at the costs of high-volume units compared with low-volume units. Watson *et al.*[92] costed NICU services using the tariffs paid to hospitals to cost high-volume NICUs compared with low-volume NICUs, and compared their effectiveness in terms of reductions in the risk of mortality to estimate the cost-effectiveness of moving £100 to high-volume NICUs. The study estimated an incremental cost per life saved of £420,000 per life-year saved.[92]

Reference costs and how they inform Healthcare Resource Groups

Historically, HRGs for infant intensive care tended to be too low and HRGs for infant special care tended to be too high. Intensive care for infants should use similar costs to intensive care for adults and so should be much higher. For example, the ratio of HRGs in 2014/15 were intensive care = 2.8 × special care, high dependency = 2 × special care, special care = transitional care. This anomaly has arisen because of the way units have submitted reference costs; there is a tendency to average the nursing over all infants rather than to apportion nurses' costs to the care needs of infants based on BAPM guidelines.[2] Differences between units' costs have also arisen as a result of the way that units apportion:

- costs between neonates and paediatrics
- costs between the different neonatal unbundled HRGs
- diagnostic costs
- layout and organisation costs, etc.

Reference cost submissions inform HRGs, but there is a lag whereby HRGs change more slowly than the reference cost submissions. Payments continue to be based on the 2001 HRGs despite the new BAPM classification in 2011. An update to BAPM 2011 was agreed in 2015, and this went through the appropriate systems to flow into the NHS data collection systems from December 2016. There are now two sets of data being collected: HRG 2001 for price and payment, and HRG 2016 for reference costs. *Figure 19* shows the information flows relative to the two BAPM classifications.

Length of stay and costs

Methods

In this section, we explore the impact of service configuration on LOS and cost the LOS using a microcosting approach based on the HRG per diem reimbursement.

Length of stay (LOS) is defined as the number of days from admission to hospital discharge or death, whichever takes place first. In our analysis, we assumed that the infant spell was censored if the last episode for an infant was a transfer to another hospital (detailed results available from the authors).

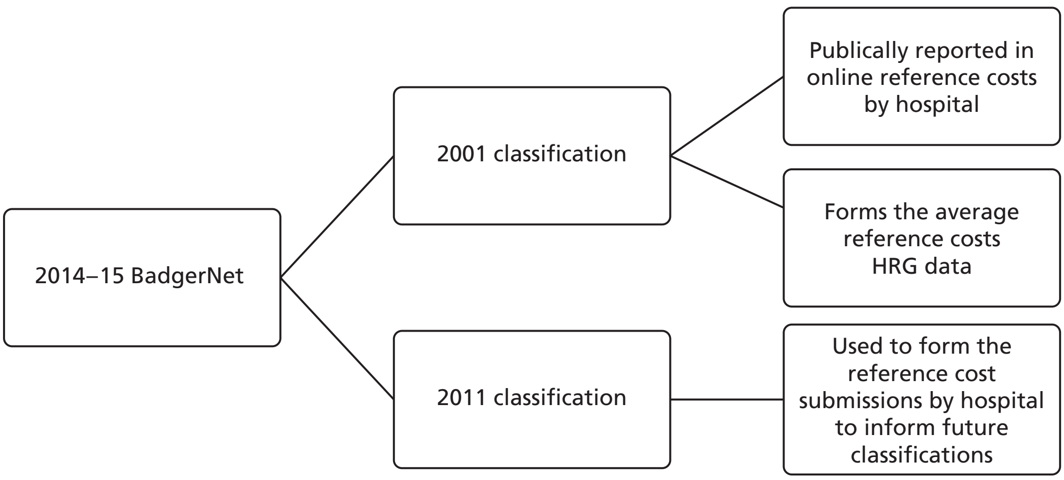


FIGURE 19 Example of information flows for 2014/15 data.

The costs of reimbursement for neonatal services for the whole inpatient spell were derived by applying HRG per diem reimbursement tariffs for 2015 based on the 2001 reference costs and payment system, which were the reimbursement opportunity cost to hospitals at the time of this study. In 2015, tariff costs were as follows: intensive care-days were reimbursed at £1176.47, high-dependency-days at £847.15, special care-days at £532.95, normal care-days at £424.35 and transitional care-days at £464.23.

We consider the impact on LOS and reimbursement of two service configurations: (1) high volume and (2) birth in a NICU compared with birth in a LNU or SCU. As with the mortality modelling, for the analysis of high-volume units we use the whole data set, whereas, for the comparison of NICUs with other levels of care (LNU and SCU), we use a slightly smaller data set that excluded infants with an incomplete number of data on the closest LNU and SCU.

The analysis used an IV approach similar to the one employed to analyse mortality. Following convention, LOS and costs were assumed to follow a log-normal distribution,[93–95] whereas the two additional equations, for the SCU and LNU binary treatment indicators, were modelled as before using a probit equation in each case. The same instruments and covariates as for the analysis of mortality were used for obtaining estimates of these models, and included covariates for gestational age, gestational age squared, infant sex, last decile of IMD score, mode of delivery and number of fetuses. We present the results of naive OLS regressions of the LOS and cost equations for comparison.

In order to avoid problems in convergence of model estimation, the model that was developed included 172 cases that presented incomplete hospital spells without adjustment for censoring (1.5% of overall data).

Results

Table 17 shows that the total LOS following birth in a high-volume unit is 9 days longer and costs £5715 more to commissioning bodies than birth in another neonatal unit.

TABLE 17 Causal effect on LOS and costs of birth in a high-volume unit ($n = 12,687$)

Estimated effect/parameter/ test statistic	LOS (days): Naive univariate OLS regression	LOS (days): IV multivariate linear model with probit treatment equations	Reimbursement costs (£): Naive univariate OLS regression	Reimbursement costs (£): IV multivariate linear model with probit treatment equations
Birth at high volume, absolute risk difference (SE)	0.6 (1.2)	9.1*** (2.7)	231 (749)	5715*** (1676)
Instrument strength: the extent to which travel time (minutes) predicts attendance at hospital, coefficient (SE)	N/A	–0.018*** (0.001)	N/A	–0.018 (0.001)***
Likelihood ratio test statistic	N/A	31.7***	N/A	31.8***
Hausman test *z*-statistic of H0: no endogeneity of birth at high-volume unit	N/A	3.74***	N/A	3.86***

*$p < 0.10$; **$p < 0.05$; ***$p < 0.01$.
H0, null hypothesis; N/A, not applicable; SE, standard error.
Notes
Log-linear estimates are back-transformed to original units adjusting for the non-linear effect on estimates of the variance. Analysis adjusts for censoring in data from 199 infants (1.6% of total sample), assuming that censoring occurs at random. Controlled covariates: age and age squared at birth, birthweight and birthweight squared, sex, deprivation of residence, mode of delivery and fetus number.

Table 18 shows that the mean total LOS following birth in a LNU is shorter by 1–2 days, whereas birth in a SCU results in 3–4 fewer total hospital days, relative to a NICU. Although the IV estimates are not significant, the diagnostic test results are consistent with the idea that the variables of interest, birth at LNU and birth at NICU, are not endogenous and, therefore, a simple OLS model may provide valid approximation to the true effects on LOS. The same result applies to IV estimates of effect on reimbursement costs, which were both statistically insignificant. A simpler OLS model suggests that a birth in a SCU is £1870 less costly to commissioning bodies than a birth in a NICU, although the effects are imprecisely estimated. In contrast, reimbursement costs for a birth in a LNU is £643 less costly to commissioning bodies than a birth in a NICU, but the result is not significant. These log-linear model estimates are back-transformed to original units, adjusting for the non-linear effect of the error variance on treatment effect estimates.

TABLE 18 Causal effect on LOS and costs of birth in lower-level hospitals (LNU and SCU) relative to NICU ($n = 11{,}037$)

	LOS (days)		Reimbursement costs (£)	
Outcome	Naive univariate OLS regression	IV multivariate linear model with probit treatment equations	Naive univariate OLS regression	IV multivariate linear model with probit treatment equations
Birth at LNU, absolute risk difference (SE)	–1.9* (1.1)	–1.4 (1.9)	–643 (681)	834 (1180)
Birth at SCU, absolute risk difference (SE)	–3.5** (1.7)	–2.7 (2.9)	–1870** (1042)	–1770 (1772)
Instrument strength for minimum travel time (minutes) to NICU, absolute risk difference (SE)	N/A	LNU equation 0.033 (0.001)*** SCU equation 0.015 (0.001)***	N/A	LNU equation 0.033 (0.001)*** SCU equation 0.015 (0.001)***
Instrument strength for minimum travel time (minutes) to LNU, absolute risk difference (SE)	N/A	LNU equation –0.054 (0.001)*** SCU equation 0.007 (0.001)***	N/A	LNU equation –0.054 (0.001)*** SCU equation 0.007 (0.001)***
Instrument strength for minimum travel time (minutes) to SCU, absolute risk difference (SE)	N/A	LNU equation 0.007 (0.001)*** SCU equation –0.062 (0.002)***	N/A	LNU equation 0.007 (0.001)*** SCU equation –0.062 (0.002)***
Likelihood ratio test statistic	N/A	46.04***	N/A	45.9***
Hausman test *z*-statistic of H0: no endogeneity LNU treatment variable	N/A	0.26	N/A	1.57
Hausman test *z*-statistic of H0: no endogeneity SCU treatment variable	N/A	0.29	N/A	0.03
Test *z*-statistic of H0: valid overidentifying restriction of minimum travel time to NICU	N/A	0.40	N/A	0.36

*$p < 0.10$; **$p < 0.05$; ***$p < 0.01$.
H0, null hypothesis; N/A, not applicable; SE, standard error.

Notes

Controlled covariates: age and age squared at birth, birthweight and birthweight squared, sex, deprivation of residence, mode of delivery and fetus number.
Log-linear estimates are back-transformed to original units, adjusting for the non-linear effect on estimates of the variance.
Analysis did not adjust for censoring in data from 199 infants (1.6% of total sample).
Analysed excluded data on infants who had incomplete IV data on travel time to closest LNU or SCU: 1650 out of the original 2687 infants (13%).

Discussion

Length of stay following birth in a high-volume unit has a mean duration of 9 days longer and a mean cost of £5715 more to commissioning bodies than LOS following birth in another neonatal unit. LOS following birth in a LNU is shorter by 1–2 days, whereas birth in a SCU results in 3–4 fewer total hospital days, relative to a NICU. For reimbursement costs, a birth in a SCU is £1770 less costly to commissioning bodies than a birth in a NICU, although the effects are imprecisely estimated (with overlapping CIs). The reimbursement costs to commissioning bodies for births in a LNU are no different from those for births in a NICU. This appears paradoxical given the longer total LOS for birth in a LNU; however, a possible explanation for these results is the different production functions between NICUs and LNUs in terms of their relative use of number of days at different levels of care.

We will return to the estimates of LOS and reimbursement again in *Chapter 9*, *Evaluation of high-volume neonatal intensive care units compared with other hospitals*, when we calculate the effectiveness of high-volume units and NICUs.

Parent costs

Data

Data were collected by BLISS on 1347 parents for neonatal events between 2010 and 2014 in the UK for infants with a gestational age of between 25 and 34 weeks, most of which took place in England (89%). The questionnaire collected clinical information about the pregnancy, infants and place of birth (gestational weeks, hospital and unit type, infant additional hospitalisation and LOS). Information was also collected on the financial status of individuals during the period when the infant was born and the expenses paid by the parents during the visit to the neonatal unit (overnight stays and the relative cost, costs of parking, food, public transport tickets, travel and childcare, how neonatal hospitalisation affected the family budget and access to new loans). Finally, information was collected on sociodemographic status (such as income, sex, age, relationship status and ethnic group).

Methods

An OLS cost model was developed in order to capture the factors that define and influence the costs borne by families during the event of a birth in a neonatal unit. These costs are considered ‘out of pocket’, meaning that they are not supported by the NHS, but they can have a significant impact on family budget, especially considering the long LOS of preterm infants in neonatal units.

The model evaluated the feasibility of a regression model in relation to several variables or characteristics of infants and families using a linear regression model. The dependent variable (the total costs for families) was defined by the following covariates: cost for food and travel (in GBP), the use of childcare and baby care when parents were away, overnights spent, the purchase of breast pumps, the use of parking, the use of unpaid leave, a dummy variable that represents parents both having had unpaid leave, if the employer of the partner was supportive in the maternity period, the average household income of parents, the days of visit per week in the neonatal unit, the presence of children at home and the relative number, the distance in miles from the birth hospital, the age of the parent, the use of public transport, the use of private and public transport and if the parent is in a couple.

Results

At an early stage of the analysis, concerns were raised over the missing data. Some answers were mostly complete; for example, infant LOS was only missing for 6% of cases. Other missing answers are attributable to families finding it difficult to recall such information; for example, the distance in miles (29% of cases were missing) and time (81% of cases were missing). The questionnaire contained 100 questions, with 22 questions relating to financial information, and some open-text questions. The final model is based on 614 complete observations, so caution is needed regarding the generalisability of these results.

Table 19 summarises the main results and shows that the cost of food and travel, the use of baby care, the use of car parking, the unpaid leave and the presence of unpaid leave for both parents, the average income and the support of the employer of the partner during the maternity period are all significant. Factors that reduce costs are the partner's employer (a higher availability of the partner can help to reduce parents' expenses), the LOS of the infant, and if the mother is in a couple (even if all the covariates are not significant). The model shows a good fit to the data with an adjusted R^2 of 0.5847.

Discussion

The model shows that unpaid leave, food, travel, baby care and parking all have a significant impact on costs. The support from a partner's employer can reduce costs, as does the availability of the partner to help (e.g. with preparation of meals to take to the hospital and other facilities).

The questionnaire was long, which may have increased the likelihood of missing data. To improve completeness, we recommend fewer questions that did not use free text.

TABLE 19 Family costs regression model

Family cost	Coefficient	Standard error	*t*-value	*p*-value	95% CI
Food***	1.260	0.277	4.550	0.000	0.717 to 1.804
Travel***	1.069	0.125	8.570	0.000	0.824 to 1.313
Use of childcare	3.548	30.633	0.120	0.908	–56.615 to 63.711
Use of baby care***	53.622	18.435	2.910	0.004	17.416 to 89.827
Use of overnight stay	25.758	50.885	0.510	0.613	–74.179 to 125.695
Purchase and use of breast pump	28.238	131.118	0.220	0.830	–229.274 to 285.750
Use of car parking***	63.358	19.474	3.250	0.001	25.111 to 101.606
Unpaid leave***	484.903	20.517	23.630	0.000	444.607 to 525.199
Unpaid leave for both parents***	228.359	47.345	4.820	0.000	135.375 to 321.343
Support from partner's employer**	–42.203	19.859	–2.130	0.034	–81.206 to –3.201
Household income***	0.002	0.000	3.660	0.000	0.001 to 0.000
Number of days of visit in unit per week	7.525	8.192	0.920	0.359	–8.564 to 23.614
Children at home	9.601	11.147	0.860	0.389	–12.292 to 31.494
Distance in miles from birth hospital	0.037	0.318	0.120	0.908	–0.588 to 0.661
Age	2.445	1.779	1.370	0.170	–1.050 to 5.940
Use of benefits	22.537	23.512	0.960	0.338	–23.641 to 68.714
Use of only public transport	16.565	48.250	0.340	0.731	–78.198 to 111.327
Use of public and private transport	31.75	22.81	1.39	0.16	–13.04 to 76.55
Couple status	–23.07	36.86	–0.63	0.53	–95.46 to 49.32
Weeks of gestational age	0.11	2.77	0.04	0.97	–5.33 to 5.55
Weeks of LOS	–1.92	1.48	–1.30	0.20	–4.84 to 0.99
Constant	–204.12	130.68	–1.56	0.12	–460.76 to 52.52

**$p < 0.10$, ***$p < 0.05$.

What is important to families?

The objective of this part of the study is to undertake qualitative research on the factors that families and policy-makers would like to see taken into consideration when determining service reconfiguration. The aim is then to assess the feasibility of including these aspects in a DCE, which is typically used in health economic evaluations.

The interest in DCEs in health-care decision-making has increased in recent years. DCE is a method that allows a number of characteristics to be traded off against each another.[96,97] Janus *et al.*[97] report a systematic review of preference elicitation studies, defining categories for studies that informed clinical decision-making, supported reimbursement decisions (as in health technology assessments) or elicited the perceived benefits and risks of health innovation for the market authorisation of drugs; in all of these types of decision, DCEs were adopted.

There are review articles summarising how qualitative research can be used to inform health-care research.[98–100] There are also applied examples of how qualitative research has been used to inform outcomes important to families and policy-makers in other areas (e.g. for children and young people with neurodisability).[101] In addition, we are aware of two methodological papers that give detailed advice about how attributes might be developed for a DCE,[102,103] but very few DCEs report the attribute development stage in any detail.[104] In this research, we follow the steps suggested by Coast *et al.*[102] and, like Klojgaard *et al.*,[103] we recommend cognitive interviews to help identify if any attribute might prove problematic in the DCE. Data collection may take the form of interviews or focus groups in the absence of a well-constructed meta-ethnography. Coast *et al.*[102] suggest that the choice between these methods may be a result of practicalities: interviews are recommended for sensitive topics (attitudes towards end of life) and focus groups are recommended if discussion among those affected may reveal additional issues.

The preferences present in DCE studies are related to health and non-health outcomes, processes and service characteristics.[104] A crucial element in the DCE process is the selection of appropriate attributes for outcomes and process characteristics. It is also likely that including qualitatively different attributes can also increase the complexity of the decision and may make the choices harder to complete. If we consider service reorganisation, such as centralising health services, the impacts can be present in both health, non-health and process characteristics.[105] DCEs have been proposed to examine patients' and decisions-makers' preferences towards such reorganisations of care, such as those currently under review for centralisation of neonatal care.[33] However, such applications face the additional challenge that the outcomes may take place at different points along the care pathway (e.g. risk of hospital mortality for the mother or child vs. risk of longer-term childhood disability) that increase decision complexity. Although no formal DCE study been undertaken here, the qualitative research will be used to inform the outcomes of interest that will be used in a later DCE and to assess the feasibility of using DCEs in future service reorganisations of neonatal care.

Systematic review

The current guidance for attribute selection in DCEs emphasises the need to achieve a balance between the competing objectives of the participants and the decision-maker, the relevance of the research question(s) and if attributes are related to one another.[106–108] However, the challenges faced in selecting appropriate attributes for service provision, outcome or both have received less attention. The systematic review examines the extent to which researchers investigating antenatal and neonatal care through DCEs consider and justify in their design whether the attributes are for service provision, outcome or both (see *Report Supplementary Material 1*).

Methods

Systematic electronic searches of prespecified terms were performed in EconLit, EMBASE HMIC (Healthcare Management Information Consortium), MEDLINE, NHS EED (Economic Evaluation Database), PsycINFO and Web of Science databases, from 2000 to June 2016. DCE studies investigating topics on premature infants, neonates, newborns, or mothers/fathers/parents were included. Studies were excluded if the participants were children or adolescents aged < 16 years.

Results

After removing duplicates, 9701 unique results were identified but only 299 DCEs for antenatal and neonatal care were initially considered, of which 13 met the inclusion criteria. The selection of studies is presented in the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram[109] (see *Appendix 3*, *Figure 31*).

Most of the studies were conducted in industrialised countries (UK, $n = 4$;[110–113] Australia, $n = 3$;[114–116] the Netherlands, $n = 2$;[117,118] and Canada, $n = 1$[119]), and three were conducted in developing countries.[120–122]

The objective of the elicitation exercise varied in all studies from types of obstetric services,[117] screening tests for Down syndrome,[113] induction of labour[114] and delivery,[118] perinatal experiences[119] and supplementary diet,[121] among others. Most of the studies involved women participants, and health-care providers were included in only two studies.[115,122] Almost 80% (10/13) of the studies focused on antenatal care, including one that considered both antenatal and postnatal care. The number of individuals participating in the DCEs varied from 56 to 1464 (mean 284, median 130). The mode of administration was also diverse, with web-based questionnaires;[110,115,118] postal questionnaires,[111,117] and pen-and-paper questionnaires at clinics and teaching hospitals,[113,114,119,121] with the rest reporting using a questionnaire but without specifying the mode of administration.[112,116,120,121] The reduction in studies selected (from 299 to 27) due to eligibility is because the vast majority related to paediatric rather than neonatal care and/or did not relate directly to DCE. Overall, the studies showed a heterogeneous picture of selection of attributes for service provision, outcomes or a mix of both, but there is a lack of consideration or justification for these choices.

Summary of approaches used to develop attributes within these discrete choice experiments

Attributes were selected by reviewing existing studies,[111] as part of the characteristics of the intervention being evaluated in a randomised controlled trial,[113,118] qualitative interviews with patients and/or stakeholders,[112,113,117,118,122–124] literature review and qualitative interviews,[110,115,116,120,125–127] and international guidelines.[121] However, in some studies it was unclear how the authors reduced the number of attributes obtained in qualitative research to a manageable set of attributes.[116]

Focus group

At the beginning of the study, we started with a set of attributes suggested by the pilot work, which included one process outcome (access to neonatal services) and three health outcomes: maternal mental health, infant death and risk of childhood health problems. We presented these attributes to the first focus group before conducting the qualitative interviews, giving examples of how they might be shown to parents:

- maternal mental health – reduce anxiety and depression
- risk of infant death for those born at 24 weeks of gestational age – reduce from 5 in 1000 to 2 in 1000 infants
- risk of childhood health problems – eyesight problems increase from 1–2 in 100 to 2–5 in 100 infants
- access to neonatal services – increases travel time for some families from 60 to 120 minutes.

Although some of the outcomes, like access and travel times, are more straightforward, the issues of risk and uncertainties over morbidity are clearly complex, and so the research set out to explore the feasibility of DCEs, or other approaches, to collect data to inform the weights that decision-makers can apply in this context. Examples of framing considered for uncertainty in focus groups were as follows:

- Low confidence – our confidence in this effect estimate is limited.
- Medium confidence – we are moderately confident in this effect estimate.
- High confidence – we are very confident that the true effect lies close to our estimate.

Or:

- Between 1 and 10 infants, most probably 5 infants, out of 100 will experience eyesight problems.

Most parents felt that the presentation of these options was difficult and not easy to understand and compare, and two parents preferred to use the confidence levels rather than the values reported for risks. We also presented parents with a table to illustrate risk with a pictogram with 100 faces, showing 2% chance as two faces of different colours. Generally, it was felt to be a clear presentation of risk. We also showed parents the protocols we were aiming to use in the qualitative study, which aimed to discuss the parents' experiences focusing on a set of open questions. Little additional feedback was given on these protocols in the focus group.

Patient preference interview

The qualitative study built on earlier public and patient involvement work,[33] which aimed to identify outcomes that were important to families of children requiring neonatal care. The qualitative research explored these outcomes further and aimed to refine the language that is used to describe these outcomes, to explore in what ways these outcomes are perceived to vary by parents and to identify if any attribute might prove problematic in the DCE.

Methods

Recruitment of participants

We purposively sought to interview parents who had a child discharged from the neonatal service within the previous 6 months to 5 years, who were interested and willing to share their experience of using the neonatal service. We aimed to recruit a varied range of parents to provide further insight into the issues and outcomes that are important and how they describe those outcomes. Carrying out the interviews at participants' houses broadened the scope of who we could interview.

Neonatal support groups, such as BLISS and SNUG (Supporting Neonatal Users and Graduates), hold a list of parents who have consented to be contacted for future research. We advertised the study through this list of parents and through a conference in south-west England on neonatal services to parents held in October 2016 in Torquay, as well as through families known to the neonatal unit at Royal Devon and Exeter NHS Trust through the lead link, Sue Prosser, who is Matron of the neonatal unit at Royal Devon and Exeter NHS Trust. The advertisement is provided in *Report Supplementary Material 2*.

At the recruitment stage, prospective participants were provided with written information about the purpose of the research, what the interviews would involve, how long the interviews would take and how their data will be used (see *Report Supplementary Material* 2). Prior to the beginning of the interview, participants were asked to sign a consent form (see *Report Supplementary Material 2*).

Flexible topic guide

A flexible topic guide for individual interviews with parents (see *Report Supplementary Material 2*) was developed. The semistructured probing questions were based on a series of attributes that were raised from patient and public involvement (PPI) in a previous study of neonatal care;[33] a review of the literature; outcomes suggested by the National Perinatal Epidemiology Unit (www.npeu.ox.ac.uk/research; accessed January 2018); and PPI feedback from a NeoNet PPI group held in Exeter in July 2016. In 2017, a set of outcomes for neonatal care was under development for the COMET initiative (www.comet-initiative.org/; accessed January 2018),[128] but the study was at too early a stage to inform our work. The topic guide was piloted with two parents and then amended in the light of feedback prior to commencing data collection.

Interview process

The 10 interviews were carried out by Katie Kelsey, five in the University of Exeter St Luke's Campus, four in family homes and one in a children's centre following a SNUG coffee morning. The lengths of the interviews were tailored to suit the participants, and varied from 35 minutes to 2 hours. Both parents were interviewed in two cases, and the mother was interviewed in all other cases.

Data management and analysis

The interviews were recorded digitally and transcribed verbatim. During transcription, all names and places were anonymised: any names of parents reported here are pseudonyms. Thematic analysis supported through a framework approach was used.[99,129]

Two researchers (KK and PL) initially read through the transcripts and developed a thematic coding framework that captured the factors that parents would like to see taken into consideration in determining the configuration of neonatal services and how they describe them. The coding framework was then used to code the transcripts to generate relevant themes and subthemes using NVivo version 11 software (QSR International, Warrington, UK). The framework (see *Report Supplementary Material 2*) was developed further as they (researchers KK and PL) became more familiar with the content.

Katie Kelsey and Paolo Landa separately coded two transcripts to check for comprehensiveness and consistency of coding. Differences that arose in interpretation between the researchers were discussed. Katie Kelsey and Paolo Landa then coded all remaining materials from the interviews.

Thematic charts were developed:

- Thematic matrices were created in NVivo version 11 to help with the analysis.
- The coded data for each transcript were summarised in the matrices so that for every theme and subtheme we had a summary of each person's related dialogue.
- Each of the themes were then synthesised further so that we had a condensed version of each theme.

The summaries for each of the main themes were presented to a final PPI group meeting on 5 June 2017, to which all of the interviewed parents were invited. This group validated the results and discussions took place to further refine the outcomes.

Ethics approval was granted by the University of Exeter Medical School Research Ethics Committee (reference Dec16/B/096Δ2).

Results

Ten families were interviewed using the flexible topic guide (see *Report Supplementary Material 2*), which explored the following characteristics:

- Hospital environment.
- Travel time and means of transport.
- Impact on the family (emotional and financial).
- What is important for the family?
- Language used, sensitivity, medical language, how parents explain the events, services and treatment.
- Understanding of risks – infant survival and long-term disabilities.
- Mitigating aspects – family support, SNUG and BLISS support and hospital staff support.
- Background – gestation weeks, LOS and other children in family.

The families that were interviewed all had very different stories. The gestational time ranged from 24 to 34 weeks and the LOS ranged from 2 to 17 weeks. For half of the families, this was their first child. Some of the births were in a NICU and some were in a LNU; some of the babies born in a LNU were transferred to a NICU.

A summary of the synthesis under each theme as presented to the PPI group is presented in the following sections. We identified the themes in the coding process (see *Report Supplementary Material 2*).

Hospital environment

The families that were interviewed reported having very profound experiences in the neonatal units. They reported feeling deep and contrasting emotions of trauma, anxiety, frustration and excitement as they lived through the first weeks of their baby's life, often not knowing if their baby would survive. One mother spoke of the moment of excitement when the hospital staff began referring to the time 'when' she would take her baby home, implying the 'if' prior to that. They all had very vivid and detailed memories of their time in the neonatal units, even years after their baby was born. The parents became attached to the care teams/nurses and felt that they were 'like family'. Communication was very important and parents felt supported by the staff and that they could always ask questions. They would frequently telephone for updates, and felt that their baby was getting the best care.

There were also some occasions when parents noticed a problem with their baby and they had felt that it took a long time for the hospital staff to take notice.

Parents found it very helpful to have a tour of the neonatal unit before the birth, as they were then mentally prepared for the sight of their baby in an incubator and attached to tubes and monitors. An issue highlighted by some parents was the difficulty they found with initial bonding. This was for various reasons: partly because of all the equipment, partly that there were so many other people involved in the care of their baby and also the anxiety around the baby's vulnerability. It made a big difference when the hospital staff encouraged them to be involved.

The understandable attachment parents felt towards their care teams meant that transferring between hospitals could be a source of anxiety, even when moving nearer home to a lower level of care. On the other hand, those who had other children and were travelling every day to the neonatal unit and managing other childcare found that moving nearer to home was of great benefit. Some parents found the atmosphere in the NICU to be intense and preferred the more hands-on nature of the LNU.

Some families/mothers made good long-term friends while in the neonatal unit, as these were people with whom they had shared an extreme experience.

It was very important to families that they be involved in the care of their baby. If parents were travelling to the unit every day, and/or juggling other childcare, it made a big difference when the hospital staff took into account their timings so that they did not miss their baby being fed or washed.

It has been reported in recent studies[130–132] that it is important to involve parents in the process of care when their infant is in a neonatal unit. This process usually consists of feeding the infant with breastmilk, cleaning and bathing, taking care of the infant and letting the infant feel the mother's presence.

Some parents felt that it was crucial for them to have been able to stay at the neonatal unit. Those who did not stay spent every day there and found that the facilities made a big difference. It was important that they could bring their other children to the unit, and the nurses made this very possible:

> *[. . .] although it was a stressful, worrying situation you're in, it was nice at the same time, it was really bizarre, it was . . . It's just like another family, you know [. . .]*
>
> *Family 3 d*

> *[. . .] I think it's just, trying to be there for your baby, because it's such a strange sensation, because when you have a normal birth and you take your baby home straight away, there's obviously that bonding that happens immediately. Whereas, with a premature birth, the doctors and nurses have all that first contact, so particularly when you first come on to the unit and you're around your baby, you're not really sure what you're allowed to do or what you should be doing and, you know, when*

> *we first came on to the unit, I just didn't think I'd be able to touch him or have any contact with him at all [. . .]*
>
> Family 6 m

Impact of travel

It should be noted that parents are willing to travel anywhere and make whatever sacrifices necessary to help make sure that their infant has the best care possible. Sometimes this is at the expense of the rest of the family. Some parents were travelling by car for ≥ 1 hour each way every day to the neonatal unit. Those parents without cars relied on public transport and family members, and in some cases spent ≥ 3 hours per day travelling. The impact of travel becomes much more pronounced when there are other children in the family, with the parents trying to fit their time at the neonatal unit around school times.

Parents can feel torn between their children at home and the need to look after their baby as much as possible, and they can feel guilty as soon as they leave the hospital. They had been advised by health-care professionals that the older children will remember their lack of attention whereas the baby will not, so they tried to make sure that they included the other children as much as they could:

> *[. . .] And all of this has a cost involved in it, which is irrelevant, really, because – well, it is relevant, but you're gonna do everything you can for your child, for your baby, you know [. . .]*
>
> Family 2 d

> *[. . .] I would spend, like, a Sunday night, so I could get all her school uniform ready at home and a Wednesday night at home just so it broke the week up for her [. . .]*
>
> Family 7 m

> *[. . .] that was the toughest time. That was the hardest time, being away from everyone, not having any support [. . .]*
>
> Family 4 m

Family disruption

Families are disrupted in different ways and some of the effects are felt for years after the birth. When the mother and baby are in hospital, the separation is felt by the rest of the family. Parents report struggling with difficult behaviour because other children feel neglected and/or are worried about their sibling.

Families spoke about living in bubble away from family, in which parents, especially the mother, is focused only on the baby's health and development. For parents, it is a stressful and traumatic time affecting the whole family. Parents feel guilty about leaving their baby behind and also feel guilty about neglecting their other children.

Strains are felt between parents who feel that they are on an emotional 'rollercoaster', with each parent feeling the strain differently and trying to stay strong for the other. When fathers had to go home, leaving mothers in hospital, this created other tensions and anxieties.

The wider family was also affected as people do not know what to say or how to act. Parents felt that:

> *[. . .] every day we ended up saying sorry [to each other] and starting each day [. . .]*
>
> Family 2 m

> *[. . .] They came in and saw [baby] all hooked up to god knows what else, you're trying to explain to them that everything's OK, but they're like, 'well, come on mum, he's got, like, a tube down his throat, he's got one in his tummy . . . he's got a cannula in and he's . . .', you need someone to*

explain to your children, as well, . . . this is fine, . . . this is helping . . . it affects everybody, definitely, my mum and my dad [. . .]

Family 1 m

Information/language used

Parents would try to be in the unit for the doctors' rounds and would ask nurses afterwards if they did not understand what the doctor had said. They felt that the doctors were good at explaining the information in lay terms. Parents seemed to learn the medical terminology very quickly so that they could follow what was going on.

Mostly, people would appreciate the practical information, in stages, so that they could be involved and know what to do, only having the information they needed for that day.

It took a bit of getting used to the language:

You hear the worst things – . . . 'brain bleeds', but then they tell you it's nothing to be worried about, it's 'like a bruise'.

Family 9 m

Parents liked to be given information directly/bluntly, without health-care staff 'beating round the bush'; they reported always wanting to know the situation, and not to be told that everything is fine. They felt that the neonatal unit was better than the maternity ward at providing information:

[. . .] I think they were pussyfooting around it a little bit too much, maybe . . . they were sort of indirectly telling us what was gonna happen or what would happen if we didn't do this or didn't do that, and I think if they'd just said 'Look, if she stops growing there'll be serious problems, so we've got to get her out, basically [. . .]

Family 3 d

[. . .] Doctor [name removed] was very good and talked to my husband . . . until his questions finished and he had resolutions or some kind of answers . . . the fact that he had been heard was really, really important. You know, you can't always give an answer or solve the problem but at least he'd been heard [. . .]

Family 5 m

Understanding risks

Parents found discussions about risk difficult to process, particularly because they were immersed in such an emotionally charged experience, and they liked to focus on what they could do.

The possibility of their baby not surviving was always on their minds. For most parents, there had been some sort of discussion between them and the medical staff about the risks. Some people wanted more information than others. Mostly, parents wanted to know what might happen, but not necessarily the percentages (or likelihood).

In some cases, there were discussions about the potential risks of certain treatments (e.g. loss of use of limb when arterial line was put in, but it was to give their baby a better chance so there was no choice). Risks were also discussed in other cases in which the infant was transported by plane or ambulance. When there was a treatment or procedure with more than one possibility, parents tended to defer to the medical expertise.

Parents felt that they were given hope, but that it was realistic. Some felt that discussion was too broad and not specific enough to their case.

They used expressions like 'if things went the other way' and 'still touch and go' when referring to the condition of their baby:

> *[. . .] one of the neonatal doctors came to see me when I was in the labour ward . . . told me that he might not survive. They've got to tell you the pros and cons haven't they? I'm just telling you [name removed], he might not survive, he's very, very early [. . .]*
>
> *Family 10 m*

> *[. . .] we knew the risks and we know we have a long battle . . . But it was always done in a nice way that there was always hope, which was nice [. . .]*
>
> *Family 2 m*

> *[. . .] her chances of survival, we didn't want to know that [. . .]*
>
> *Family 8 m*

> *[. . .] if something was gonna happen now which would affect in those few weeks, then just tell us straight . . . say – if we don't do this now, in 3 or 4 weeks this could happen. Brilliant, that's . . . direct [. . .]*
>
> *Family 3 d*

Mitigating aspects

We looked at the factors that parents felt had helped them. These are summarised in the following list:

- Confidence in the hospital staff. Parents felt well supported by the hospital staff. In some cases, they had a care team and always knew that they could talk to any member of the team.
- Being able to stay at the neonatal unit.
- Being hands-on with their baby. They were able to and were encouraged to be involved in all aspects of their baby's care. When parents were travelling to the neonatal unit each day, the unit staff would mostly try to hold back the feeding/washing times and doctors' rounds until they got there.
- Thoughtful practices, such as when a mother had to be transferred to the NICU after her baby and staff had prepared a room and taken photos of her baby, so that she arrived to a lovely welcome.
- Playrooms for other children, so that they could come to the unit too and not be left out of the picture.
- Families rallying together, helping with childcare and providing emotional support.
- Supportive employers. Some fathers' employers had given them extra leave, and all had been understanding.
- SNUG and BLISS – parents were aware of them during their stay and had accessed information leaflets; however, most parents had contacted them once they had gone home and were hit ('hit me like a tonne of bricks') by both the trauma of what had just happened and the vulnerability of being at home without all the monitors and medical staff. Parents reported continuing their link with SNUG for years afterwards.
- Financial support from charities and children's centres.
- Friends made in the neonatal unit.

> *[. . .] The nurses are like your counsellors there, you know, they're the people that are just always there and listening, aren't they? [. . .]*
>
> *Family 8 m*

> *[. . .] I sent a message to the SNUG people, I said 'Yeah, I actually need someone and [SNUG representative] called me immediately, it was really nice and she told me to just talk through what happened – your journey and I was telling her the whole journey [. . .]*
>
> *Family 9 m*

> *[. . .] that was my precious bit, that's what I could do for my baby, is get her dressed and try and feed her [. . .]*
>
> *Family 7 m*

Summary

What was most important to the parents is that their baby had the best health outcome possible, and they were willing to do whatever they could do to help make that happen. The sacrifices made by parents can be disruptive to the family both emotionally and financially.

There was variation in the parents' preferences regarding staying in the neonatal unit or travelling to the unit each day. This did not depend on whether or not the baby was their first child. Some who had other children chose to stay in the unit if they had family support at home, others travelled to the unit each day to fit around childcare. Some parents for whom this was their first child chose to travel to the unit each day.

Patients also differed in their preferences towards feeding their babies. In one case, the parents did not want to feed their baby through a tube but instead wanted to wait until they could feed their baby with a bottle or breastfeed; other parents preferred to be involved in all aspects of the care.

In order to help understand how the results of the qualitative research can inform the feasibility of and attributes for a DCE, we have broken down the overall care picture into three components:

1. best care for the baby – this refers to/includes the medical care team, medical facilities and the health of the mother, including emotional support from family and friends
2. communication – including parents knowing what is happening to their baby, understanding what might happen (risks), what parents can do 'now', how they can prepare for the future (short and long term)
3. family involvement – including the care of the baby (washing, dressing and feeding), facilities for parents to stay, facilities for other children and preparing to take their baby home.

We suggest that parents would be unlikely to consider any attributes that compromise the first component; however, parents' preferences and circumstances vary enough in components 2 and 3 that we could perhaps develop attributes around communication and family involvement.

Interviews with policy-makers

We conducted a series of 10 interviews with policy-makers, clinicians and staff to assess their views on outcomes of neonatal care, how they might prioritise outcomes and which types of economic measures might be useful for planning. This involved interviewing members of the Neonatal Critical Care Clinical Reference Group as well as specialist commissioning groups (NHS England) and representatives of the Maternity and Children's Services Strategic Networks. In addition, the interviews were used to check the approach to costing neonatal services, and included questions on the factors that made up the reference cost submissions, how centralisation affects total costs and the facilities that should be provided for parents to mitigate some of the negative consequences of centralisation.

A recurring theme in the policy interviews was the importance of the well-being and health of the baby. Secondary issues were the availability of staff to cover the rotas and the difficulty of recruiting and retaining staff; for example, it became clear that in some areas, even though it may be optimal for parents to relocate to a neonatal unit, it was felt to be infeasible to staff such new centres.

Ten policy interviews were carried out by Paolo Landa and Anne Spencer. For those working within the neonatal units, we used the interviews to explore service-level issues that we may need to take into consideration when using financial and BAPM returns, such as staff composition and duties and exploring how infants were prioritised within the system when there were staff shortages. For those who were responsible for those co-ordinating and managing network services, we explored their role and the challenges arising from service reorganisation. For those working to support families with neonatal infants, we explored how their work helped to shape the neonatal service and the factors that they felt needed to be taken into consideration in service reorganisations. The second part of all of the interviews explored what policy-makers would like to take into consideration when determining service reorganisations.

A topic guide was developed and adapted as the interviews progressed (see *Report Supplementary Material 2*). All interviews were recorded and notes were then taken. The interviews involved representatives of a charity that works in neonatal care, two neonatologists that work in the organisation of a local neonatal network, two consultants of NICUs, four matrons, and a neonatologist who works both for a local neonatal network and as a consultant lead for a NICU.

Neonatal charity

The neonatal charity represented the families in the neonatal care process and the parents' needs and preferences in terms of resources and organisation. The main objective of the neonatal charity was to improve health outcomes for neonates through their research, and they engaged with decision-making and the NHS to influence and improve neonatal care. The charity also provided a support service to families by providing leaflets and other sources of information (e.g. videos and multimedia) and offering helplines for psychological support. Parents needed to talk to someone who was able to listen and provide reassurance and to work as a guide in the complex neonatal care setting. In addition, the charity offered training to neonatal staff to encourage good practice.

If neonatal services were centralised, the charity noted the importance of travel distance and the need to provide facilities to reduce the costs and improve the accessibility of services for families, such as by providing accommodation, travel reimbursement, meals and free car parking. They also felt that several resources were still missing for families and were not provided by the NHS, and that these gaps increased when home care, after the infant is discharged from the neonatal unit, is taken into consideration.

Neonatal network neonatologists

The interviews with neonatal network staff were focused on the network organisation and the impact of different policies. The first problem that was reported regarding the neonatal service reconfiguration was the challenge of changing the skill mix of staff. Converting a SCU to a LNU or a LNU to a NICU can cause several difficulties in terms of attracting new staff and developing the supervisory structures, and these could take > 10 years to overcome in terms of training, education and organisation. The neonatologists also acknowledged that it was hard to model the staff composition in the unit, as this depended on several factors including local availability of a trained workforce, the size of the unit and the number of cots.

We asked the neonatologists what type of service organisation they envisaged (e.g. a NICU for each local neonatal network, which seemed to be the main working model). The network neonatologists acknowledged that centralisation would improve mortality for the VLBW infants, but that infants should be located near to home if appropriate services were available. They also acknowledged the regional differences in access to services (e.g. in South West England: in Cornwall and Devon, there is only a NICU but accessibility in terms of travel time is very different from the London area network). As a result, they were aware of the need to support those families that travelled long distances by providing overnight accommodation while their infant was in a neonatal unit. Some of these costs are covered by the NHS and charities, but it was felt that the provision was often not enough.

Neonatal unit neonatologists and matrons

Four hospitals were interviewed in order to understand the organisation in the unit, the availability of resources and the different unit configurations in terms of service delivery. In our interviews, we considered three English NICUs and one English LNU.

We started by asking questions about the organisation of the neonatal services in each area. Most of the units were inside a hospital with a large service availability in terms of diagnostic examinations, laboratory tests and consultation of specialists. One of the units represented in interviews was a women's hospital in which the availability of some resources, such as psychological support to mothers and tests, were provided by another site, and the unit contracted separate service agreements for these ancillary services. The NICU interviews raised the issue that it was sometimes difficult to discharge back to a local hospital (LNU or SCU), creating some bottlenecks and delays of neonate transfer.

We then asked questions regarding staff and staff composition and how staff members were organised when there were staff shortages. Each unit had a different composition; for example, in one unit there was a large number of advanced neonatal nurse practitioners (ANNPs), whereas in the other units there was a high availability of junior doctors. This variability in the staff configuration can create large differences in terms of staff costs. We asked if some types of infant (e.g. infants in intensive care and infants in high-dependency care) were prioritised when the workload exceeded the BAPM standard and the staff availability could not cover the number of infants in accordance with the BAPM guidelines.[2] Staff from each unit advised that there was no prioritisation of infants and that each infant is treated with the same level of priority, with staff being allocated in accordance with the BAPM guidelines[2] where possible.

When we discussed the resources needed if the services were to be centralised further, and if NICUs became larger, all interviewees raised the need to increase staffing. In some areas, this may be a challenge because of the limited availability of training staff. Most units were already not working in accordance with BAPM standards for ≥ 80% of the time, and so there were concerns about staff shortage and the ability to attract staff to new sites if the unit had to be relocated to another part of the network. In terms of facilities offered to families, unit representatives noted the need to expand the provision of accommodation for families and services if services were further centralised. They also talked about the need to make efficient use of resources.

Discussion of qualitative study

The results of the qualitative interviews show that interviewees talked more about the infant as a whole, rather than separating the risks of death and childhood health problems. This notion of a combined attribute is similar to the idea of an aggregate measure of length of quality of life used by NICE,[133] which may be more meaningful if extrapolated forward to consider the longer-term impacts on infants, and not just short-term prognoses. In addition, the families made a connection between the baby's and the mother's health, and the importance of the mother's health was considered to be equal to that of the child by the focus group.

The qualitative interviews also raised other process outcomes including communication with the families and family support.

Furthermore, the qualitative study raised questions about the ability and willingness of parents to trade off health attributes for the process attributes. Although no trade-off questions were asked in the patient interviews, it became clear that mothers were unlikely to want to sacrifice these 'core' aspects of their baby's health for improvements in process outcomes. Parents stated in interviews that once they knew that their baby was receiving the best care, they could then perhaps consider other factors. This raised questions about including a DCE with combined health and non-health outcomes, because parents would always choose the configuration that favoured the best health outcomes for the infant (lexicographic preferences). However, in configurations that maintain a high level of care but affect process outcomes, trade-offs and DCEs become more feasible.

Chapter 9 Economic evaluation

The reorganisation of health-care services requires an evaluation of the clinical benefits and costs to the health service and to the wider society. The aims of this chapter are to develop and apply a framework that can be used to capture these wider consequences of neonatal care reorganisation, based on our work in *Chapter 8*. We estimate the incremental cost-effectiveness analysis based on a comparison of (1) high-volume units and all other units, and NICUs and other unit designations, and (2) three service reconfigurations from the simulation modelling.

Evaluation of high-volume neonatal intensive care units compared with other hospitals

Methods

Our study estimates the cost-effectiveness of an intervention that leads to infants being born in high-volume units rather than in other neonatal units. We also calculate the effectiveness of NICUs versus LNUs and versus SCUs. In contrast, Watson *et al.*[92] estimated the cost-effectiveness of investing an additional £100 of resources in NICUs per day. Their approach also differs from the one taken here in the way that they estimated costs. In their approach, the costs of neonatal services were based on the HRG for each hospital, which is part of the national reference cost submission and is used to inform future HRG tariffs. They investigate the extent to which hospitals with higher HRG unit (per diem) costs have lower mortality rates. In contrast, our approach is more in line with the traditional cost-effectiveness approach in that we focus on the impact of volume on outcome and LOS and then cost the LOS using the corresponding fixed tariff HRG per diem reimbursement for the number of days spent in each level of care (intensive care, high-dependency care, special care and transitional care).

The methods used to estimate LOS are described in detail in *Chapter 8*, *Methods*. In *Chapter 9*, *Results*, we bring together the LOS and mortality estimates from these two sections to calculate the incremental cost-effectiveness ratio (ICER).

Results

Table 20 summarises the IV results for LOS and costs for (1) high-volume units compared with other units and (2) unit designation (i.e. NICUs vs. LNU and NICUs vs. SCU). *Table 20* shows that birth in a high-volume unit increased the total LOS by almost 9 days, representing an additional mean reimbursement cost to commissioning groups of £5715 relative to births in non-high-volume units. Dividing this additional cost by the reduction in neonatal mortality in high-volume units (£5715/0.012) results in a cost per neonatal life saved of £460,887. If we suppose that an infant survives for 81 years, in line with the average life expectancy at birth for the English population,[134] and they remain in full health for the entire period, then we can convert the cost per life saved into cost per life-year gained. We discount costs and effects using an approach recommended in the HM Treasury Green Book for longer-term interventions,[135] and the discount rate is 2.5% for first 30 years of life, 3% from the 31st to the 75th year of life and 2.5% for 76th to the 81st year of life. This analysis results in an ICER per life-year gained of £15,620. If data become available on the life expectancies or quality-adjusted life-years of infants admitted to neonatal units, then this figure can be further updated.

The results of the analysis for hospital designation show that birth in a NICU exposes infants to a lower risk of neonatal death than birth in a LNU (a 1.9-percentage-point risk reduction), whereas there is a statistically insignificant effect of NICUs relative to SCUs, which may be attributable to the small number of SCU cases (10%), the different type of care provided and the imprecision of the IV method. Furthermore, birth in a NICU results in an additional 1–3 total days in hospital than birth in a LNU or SCU, and higher reimbursement costs for NICUs compared with SCUs, but the effects are imprecisely estimated (with overlapping CIs).

TABLE 20 Cost-effectiveness evaluation

	Causal effect on LOS and reimbursement costs of neonatal care at a unit with ≥ 100 babies weighing < 1500 g per year			Causal (marginal) effects of birth at a NICU vs. LNU vs. SCU: IVs estimates				
Output metric	Potential outcome in high-volume units	Potential outcome in other units	Difference (95% CI)	Potential outcome if born in a NICU	Potential outcome if born in a LNU	Potential outcome if born in a SCU	NICU vs. LNU, difference (95% CI)	NICU vs. SCU, difference (95% CI)
LOS (days)[a]	74.28	65.14	9.14 (3.84 to 14.45)	67.6	66.2	64.7	1.4 (–2.4 to 5.2)	2.7 (–3.0 to 8.3)
Cost (£)[a]	48,925	43,209	5715 (2431 to 9000)	43,879	44,714	42,108	–834 (–3147 to 1479)	1770 (–1703 to 5244)
In-hospital neonatal mortality[b]	0.025	0.039	–0.012 (–0.021 to –0.0034)	0.079	0.098	0.083	–0.019 (–0.037 to –0.001)	–0.004 (–0.021 to 0.029)
ICER (£/life-year gained)			460,887				–43,096	NICU more costly, no difference in effectiveness
ICER (£/life-year gained)			15,620				–2279	NICU more costly, no difference in effectiveness

IV analysis using travel time to a high-volume unit as the instrument, adjusted for censoring because of incomplete LOS and total costs of infants whose hospital spell ended in a transfer to another hospital (1.5%).

a Modelled as a log-normal distribution, jointly with a probit treatment equation.

b Modelled as a probit distribution, jointly with probit treatment equation.

Overall, a NICU is more costly and no more effective than a SCU, whereas a NICU is more effective and cost saving when compared with a LNU with an incremental cost per neonatal life saved of –£43,096 compared with a LNU. If we suppose that an infant survives for 81 years, and again discount costs and effects, the cost per life-year saved is –£2279 for a NICU compared with a LNU; however, more investigation is needed regarding these results to check the robustness of the findings.

Discussion

Births in high-volume units compared with births in other neonatal units have an incremental cost per life saved of £460,887. Comparing this initiative with other similar initiatives that save lives suggests that the intervention is likely to be cost-effective.[92,136,137] When we compare NICUs with LNUs, we find NICUs to be cost saving (although not statistically significant) and to reduce mortality, and so NICUs are likely to be cost-effective.

We also estimated the ICER per life-year gained using a set of simplifying assumptions and a declining long-term discount rate. Births in high-volume units compared with births in other neonatal units have an ICER per life-year gained of £15,620. Currently, NICE uses a threshold range of £20,000 to £30,000 per quality-adjusted life-year gained,[133] but there have been a number of projects to further investigate this threshold.[138–140] The ranges from this study of cost per life-year gained seem to fall within these current thresholds, but future work should aim to explore the quality-adjusted life-years for neonates who survive to provide more accurate estimates.

In conclusion, the estimation of LOS analysis seems to be a useful and appropriate metric against which to explore the impact of changes in service delivery, and, given the data available, was used here to estimate the commissioner's reimbursement costs. However, we are aware that these commissioning costs are likely to be lower than the actual service delivery costs, and that there are recent initiatives within the NHS to collect data that more accurately reflect the BAPM nursing requirement for each level. There are now two sets of data being collected: HRG 2001 for price and payment, and HRG 2016 for reference costs (which are more in line with BAPM nursing requirements). Future estimates of the impact of LOS on actual costs will be greatly improved by the ability to estimate actual costs of service delivery.

This analysis also pointed to areas for future research. For example, recent literature questions the use of a threshold to assess intervention efficiency when there are economies of scale or scope,[141] an issue that is most relevant to interventions looking at the cost-effectiveness of service delivery. In addition, service delivery interventions tend to cover a wider range of costs and effects, which need to be fully reflected in the opportunity costs.[142] As we move forward in the evaluative frameworks for service delivery, it seems important that such wide considerations are taken into consideration.

Evaluation of three different service configurations

Methods

Discussions with policy-makers and PPI groups and the review of the literature led to a list of key elements that were likely to be affected by service reconfiguration. In this report, we focus on three main categories that we have data on and can cost: nursing, travel times and transfers. In the current evaluation, we were unable to incorporate childcare costs (because the numbers of parents with other dependent children were unclear), costs of doctors (because, as revealed in the site visits, there are several differences arising from staff composition: the ANNPs and junior consultants have similar tasks but different salaries and categories) and overnight stays (without further consensus when these should be provided). Food, parking and unpaid leave policies also affected the costs for families, but these are not included because they are unlikely to greatly change with service configuration.

Mortality can be estimated by regression analysis, with information on birth weight (continuous, which means we can work with any meaningful categories); sex; mode of delivery (emergency caesarean not labour induced, emergency caesarean labour induced, vaginal spontaneous or unknown); quintiles of multiple deprivation; multiple foetuses (≥ 2 vs. 1); and level of hospital of birth (NICU or other) or, alternatively, high-volume units of birth (≥ 100 infants born weighing < 1500 g per year). We do not include the mortality estimates in these provisional estimations, given concerns about how best to assess the impact of transfers of infants into the analysis.

Predictions of NHS costs are based on the level of care received and a microcosting based on WTE nurses. Unit costs for WTE nurses are based on Curtis.[38] Although each band of nurses has a range of salaries attached to it, the cost of nurses was calculated based on the average unit cost of bands 5, 6 and 7 for neonatal nurses with and without specialisation for 2014/15, which amounted to £48.50 per hour. To cost infant transfers, we used the national reference costs associated with the HRG code XA06Z, which covered Transportation in Neonatal Critical Care and amounted to £1100.97 in 2015.

The costs to families that we include in the current model are travel time and vehicle operating costs. The unit costs applied to travel time are based on the Department for Transport's non-business costs of travel.[39] Travel for non-business is based on a study estimating the willingness to pay of travellers for shorter travel times.[40] This study found that travellers gave the same value to time in all modes of transport. The Department for Transport costs business travel at a higher rate, which varies between modes of transport based on lost workplace productivity and the wages of employees who typically use that mode of transport. It is recognised that travel may also take place in work time for spouses of women who have undergone caesarean sections (and are advised not to drive in the first 6 weeks), who may take time off work to drive the mothers to the units. The cost of travel time per hour in non-working time is £5.56 and the cost of travel time per hour in working time is £31.98; here we used an average cost of £14.99 per hour. The unit cost of vehicle operating is based on motoring costs set out by the AA (Automobile Association),[143] and includes an allowance for car parking.

Results

Table 21 summarises the costs of three different service reconfiguration scenarios under no capacity constraints and in which units run at ≈80–85% of maximum capacity (based on the results of the modelling shown in *Table 12*). The current configuration represents the actual configuration in England, whereas the centralised configuration considers a large reduction of units at each level of care. An alternative configuration was designed to minimise travel distances while having all NICUs receiving ≥ 100 VLBW infants per year. *Table 21* reports the presence of no capacity constraint and the constraint of running units at 80–85% of maximum capacity. On-duty nurse recourses for each unit are set at the next whole number up from a theoretical 85% of capacity utilisation (e.g. if running at 85% of capacity is calculated as requiring 5.4 nurses present at any time, then unit resources are set to six nurses at any one time). The number of on-duty nurses is given here assuming that capacity is capped in accordance with the BAPM recommendations. If there is allowed working beyond the BAPM workload recommendations,[2] then nurse numbers will scale down proportionally.

It is clear from the figures in *Table 21* that nursing costs are the largest cost component, approximately 18 times higher than travel costs and 33 times higher than transfer costs. Nursing costs also reduce during centralisation, because of economies of scale, and so are likely to be a key driver for any observed changes in overall costs. For example, in the unconstrained system, greater centralisation (from 48 to 45 to 30 NICUs) reduces nurse costs and slightly increases transfers costs, resulting in cost reductions from a NHS perspective. Incorporation of a wider set of costs, for example travel time for families, results in families paying more in travel costs, but overall the more centralised configurations still reduce costs from a societal perspective. A similar result is found for the constrained system, although the reductions in costs are lower.

TABLE 21 The impact of service configurations on costs

Costs	Impact (£M)		
	Current configuration (45 NICUs + 78 LNUs + 38 SCUs)	Alternative configuration (48 NICUs + 78 LNUs + 35 SCUs)	Centralised configuration (30 NICUs + 30 LNUs + 30 SCUs)
No capacity constraints			
Nurses	633.04	642.39	580.36
Transfers	18.95	17.62	19.099
Travel	35.27	35.45	44.29
Total NHS costs (nurses and transfers)	651.99	660.01	599.46
Total costs (NHS costs and travel)	651.99	660.01	599.46
Units planned to run at ≈80–85% of maximum capacity			
Nurses	566.34	568.46	550.62
Transfers	49.85	48.51	34.41
Travel	48.84	46.48	50.96
Total NHS costs (nurses and transfers)	616.18	616.97	585.03
Total costs (NHS costs and travel)	665.02	663.46	635.99

Discussion

The three scenarios (current, centralised and alternative configurations) show the potential impact on neonatal costs from service reorganisation. Nursing costs are the largest cost component and are reduced by centralisation; overall, this leads to reduced costs from a NHS and societal perspective. To assess the cost-effectiveness of these scenarios, more information is needed on the impact that these service reconfigurations have on mortality. High-volume units reduce mortality, but we can only show that these benefits are accrued for those born within those units. To assess service reconfigurations more fully, future research needs to explore the impact of high-volume units on infants transferred into those units after birth.

Our analysis has a number of limitations. The current analysis identifies potential savings from service reorganisations assuming that resources will be made available. We are, however, aware that some geographic areas may already be experiencing shortages of experienced nurses and consultants, and this analysis does not take into account staff availability. We have also assumed that the changes are instantaneous, and we have not modelled the transitional costs of moving from one type of configuration to another. To minimise disruption to the service, it is likely that reconfigurations will take the form of multiple sequential steps that take place over several years. The approach we have adopted for the economic evaluation is similar to that used by NICE, which does not typically take capacity or transitional costs into account in the economic evaluations underpinning clinical guidelines. However, for implementation, it is clear that these issues have to be factored in on a case-by-case basis.

Chapter 10 Information visualisation

Communication of outputs

One key and often neglected area in applied health-care research is the need to present outputs in a clear and accessible way to a variety of audiences. In this project, a number of stakeholder groups have an interest in understanding and interpreting the work. These include health-care policy-makers, commissioners, clinicians and care workers, researchers, parental groups and individuals who are the recipients of neonatal care in England as well as the public more generally. Commonly, these separate groups will have both different informational needs and differing levels of expertise and experience in the area. Therefore, it is likely that a range of formats and media will be appropriate to cater for these varying requirements and constraints when presenting our work in different contexts to differing audiences.

Using visualisation

The field of information visualisation (sometimes referred to as data visualisation) has developed over many years in recognition of the need to present information in clear and compelling ways.[144,145] It has also reflected the changing basis of media technology, in which many new methods of communicating information have evolved in recent decades (e.g. through the internet and the use of interactive/animated computer displays).

In our study, relatively complex technical outputs and conceptual associations need to be conveyed to a wide variety of audiences and there is a clear potential to use information visualisation to facilitate communication. Although a comprehensive treatment of this subject is beyond the scope of this study, we have summarised some initial thoughts in relation to specific stakeholder groupings in the following sections.

Commissioners and policy-makers

One clear community of interest for our study is policy-makers and commissioners in health care who are keen to understand the issues and evidence supporting different options for neonatal care and to use this information to inform policy decisions as well as justify such decisions to others. Responses from our interviews with policy-makers in this study highlighted the importance of communication as key to promoting change, justifying policy initiatives and for exploring options of new models of care.

A clear need here is for relatively complex data to be conveyed in accessible formats that can be shared in a decision-making context. Such data often bear on the relationships between different (sometimes opposing) parameters of interest; for instance, the relationship between cost and outcome or geographic centralisation and localisation for alternative service delivery options. In such circumstances, the use of graphs or maps to represent a range of policy scenarios and demonstrate the modelled impact of these alternatives against key metrics can be essential.

In this context, graphical standards can be very important in supporting clear communication and understanding between groups; for example, the extensive adoption of forest plots in health-care meta-analysis and systematic reviews[146] and cost-effectiveness acceptability curves in health technology assessments[147] have greatly facilitated understanding and discourse, especially for relatively complex informational needs in policy contexts. In this project, we would point to our development of the Villeneuve chart (see *Appendix 1*) as a novel and potentially valuable graphical method of conveying important information in relation to location preferences and resilience of service centres. These charts could be applicable in a variety of contexts. The use of 'violin plots' (see *Figure 15*) demonstrates another use of information graphics to convey relatively complex aspects of the data analysis.

When maps and other graphical media are used to represent important findings, it is important to be aware of potential perceptual biases that can result. This is especially the case when interest groups may wish to promote or advocate different policy alternatives. One example is the undue prominence that can be given to rural areas in shaded heat maps. Rural areas are typically shown as overly large visual areas relative to their population. Cartograms, which scale geographic regions by variables, such as population (rather than physical area), can be used to overcome this perceptual bias,[148,149] although such representations produce an unfamiliar and highly distorted image relative to a standard map of the UK. Although we did not explore the use of cartograms, this is clearly an area of interest that could be explored in this context.

Researchers

Researchers and academics commonly need to be able to extract clear and precise technical information from a study. For this reason, relevant data should always be made available in numerical form (e.g. in formatted tables). In addition, however, graphical and visualisation tools can be important to reveal trends, patterns and relationships in outputs, especially when these are not immediately apparent from the numerical data. Descriptive analyses of large data sets can particularly benefit from such methods,[144] which often provide a useful synopsis of the key findings.

Parents and the general public

In discussion with parents in the PPI workshops (see *Chapter 11*, *The workshops*), it seemed clear that map-based presentations were most helpful when describing the various scenarios for geographic service configuration. In particular, parents could easily relate to maps that described services in their local regional area because they had direct and experiential knowledge of these services. In addition, parents readily understood narratives and examples of pathways of care because stories and case study examples can be readily understood. Line graphs and charts, for example those that describe trade-offs in centralisation versus localisation of services, often needed substantial explanation to be fully appreciated by parents. In such cases, it seems clear that careful consideration needs to be given to the mode and design of communication (e.g. the design of information presentations as well as forms for interviews and choice experiments).

For communication with the general public, it is notable that some of the publicity literature[150] makes extensive use of information graphics, such as pictograms, to convey basic statistics about neonatal care in England. This approach seems particularly appropriate to help ensure that the information is both accessible and attractive to general readers and was adopted when communicating with parents in this project for some elicitation processes (see *Chapter 8*, *Focus group*).

Importantly, the use of visualisation methods in communication with parents and the public can be valuable in eliciting information as well as in presenting information. In our study, for example, pictorial representations were utilised to identify key concerns in a preference elicitation process. Further work could be useful in determining whether or not such graphical techniques could be deployed in the presentation of options and scenarios for preference elicitation within the proposed framework.

Summary of communication requirements

When communicating research outputs, it is vitally important to consider how information is conveyed as well as what information is presented. This is especially critical when such research outputs need to be used across a range of contexts and understood by a variety of audiences.

It is clear that there is a wide range of options when deciding how best to communicate and present complex information. Options include information graphics, computer media and more traditional numerical tables and text. However, the starting point for designing effective presentations should always be a consideration of the specific needs and expertise of the target audience, as outlined in this section. This will often dictate the level of technical detail required for different levels of understanding and will often support differing 'languages of discourse' in different stakeholder groups. Also importantly, visualisation tools can be useful to 'bridge across' the different groups with an interest in the area and provide a basis for shared dialogue.

Once these needs are understood, the constraints of the formats that can be used can be taken into account and different options can be explored. For example, black and white paper-based media determine a narrower range of options relative to the interactive and animated potentials of internet-based media.

Chapter 11 Parent involvement

The aims of the PPI in this project were to:

1. ensure that the modelling work and health economics that we carried out took into account the needs and concerns of parents and families who use neonatal services
2. explore the best way to communicate our findings to parents and the public and involve them in decision-making about the design and configuration of neonatal services.

Parents were paid £50 to attend each meeting, in addition to travel and childcare expenses as required. The PPI activities were led by Andrew Gibson and Sue Prosser (research team members).

Recruitment

To maximise the diversity of our parent group, we sent flyers advertising the opportunity to be involved in this project and requesting expressions of interest to all ODN managers and ODN lead nurses in England (see *Report Supplementary Material 2*). This resulted in a number of enquiries from areas across the country, from Carlisle to Cornwall.

We wanted to involve parents with direct experience of neonatal services. However, by definition, parents with these experiences frequently face various demands and pressures in their lives. They are frequently busy caring for young children who often have complex ongoing health needs. We tried to maximise the accessibility of our involvement activities through using easily reached venues and flexible times for meetings and through offering parents alternative methods to contribute [e.g. through e-mail and Skype™ (Microsoft Corporation, Redmond, WA, USA)].

Although travel and childcare were paid for, attending meetings meant that parents spent time away from their child, the impact of which should not underestimated following a potentially traumatic neonatal experience. We maximised the accessibility of all parents' meetings by making it clear that children could attend with their parents if they felt that this was important or necessary.

Consideration was also given to our duty of care towards the parents who became involved in our work. Discussions of experiences in neonatal care might raise potentially difficult or traumatic memories. Sue Prosser ensured that all who were involved had ongoing support or were signposted to appropriate additional help as required.

Initial recruitment and contact following expressions of interest was via telephone by Sue Prosser. During these conversations, the needs of the parents and the opportunities to be involved in the project were discussed.

Following initial telephone recruitment, permissions were obtained to share e-mail addresses and contact details to enable a group conversation to take place via e-mail and other media, such as Skype.

Establishing and running the parent groups

Despite our efforts, juggling childcare and hospital appointments led to some parents deciding that they were unable to commit to being involved in the project. In the end, we established two parent groups: one based in London, consisting of three parents, and one based in Exeter, consisting of five parents. We established two groups because we wanted to ensure that a diversity of experiences of using neonatal services were represented in our work and, in particular, we wanted to ensure that parents with experience of accessing neonatal services in both rural and urban environments were adequately represented, given the differential impact that the centralisation of neonatal services might have on these two groups.

Ongoing support for the groups was essential to ensure that members were kept updated and engaged because there were often 6 months between workshops. This was achieved by regular e-mail updates. Other ongoing challenges for continuing involvement included parents being unable to attend meetings because of a child's illness or because of changes in their own lives including returning to work following a period of maternity leave or finding new employment and/or training as their lives, post a neonatal experience, moved on.

A series of five PPI workshops was carried out in three phases. In the first and second phases, two workshops were run in both Exeter and London. In the third phase, a workshop was held in Exeter and London members were able to join via Skype.

The workshops

June 2016 workshops

These initial workshops were used to introduce the parents to operational research and its potential application to the planning of neonatal services. This was done by demonstrating how a simple operational model might be generated to analyse the flow of customers through a restaurant. We then applied this line of thinking to analysing the distribution of neonatal services and illustrated this with findings from our initial study of neonatal services in South West England.[33] It became apparent that the parents in the group were willing and able to understand how this type of research might be used to plan services and how issues relevant to parents might be built in to such a model so that it could reflect a number of different parameters set by various stakeholders including the parents.

During these workshops, the parents exchanged stories/experiences of using neonatal services. This had two benefits. First, it helped the parents to quickly gel as a group. Second, it was helpful for the researchers involved to appreciate the lived experience of the parents and how this might be taken into account in their work. The issues that were raised fell into the following themes:

- Daily living. Maintaining routines such as work and siblings attending school. Getting maternity and paternity leave, finding accommodation and food if living away from home for extended periods.
- Barriers to maintaining daily living. Distance travelled to access services and LOS. The strains placed on families increased as both of these factors increased. Coping with uncertainty was difficult and there was the impact of all of these factors on physical and mental health.
- Support.
    - Practical. Provision of accommodation [location (e.g. on the ward, in the hospital or in the town)]. Financial support to help with the cost of travel, parking and food.
    - Psychological. Stress on mother and on marital relationships and impact on siblings. A need was identified for ongoing psychological support in some cases, which could be provided through peer support.

It became apparent that when considering the potential of centralising neonatal care to improve outcomes for neonates, thought needs to be given to measures to mitigate the potential negative impacts on parents and families. This is likely to have a positive impact on the longer-term health and well-being of the neonate.

These workshops also highlighted the different experiences of living in semirural locations compared with urban locations. Cities potentially offer good public transport links. For people in rural areas, using public transport can greatly increase travel times. Finding accommodation for overnight stays within a hospital may also be more difficult in some urban areas and older hospitals.

Our health economics team also presented a planned DCE to the group. The parents raised concerns about some of the terminology used and the assumptions made: for example, that morbidity had been explained as 'your baby having eye problems'. The group suggested that the health economics team needed to review its approach to ascertaining the 'cost' of using neonatal care (see *Chapter 8*, *Results*). Eventually, it was decided to employ a qualitative researcher to carry out interviews with a sample of parents to develop this further.

November 2016 workshops

We held a second pair of workshops in November 2016 in both Exeter and London. Parents were presented with the initial findings from the study. We explored with the parents how best to present differing outputs from the operational research. This included the use of graphs, maps and Pareto fronts. The parents found maps and Pareto fronts very visual and relatively easy ways to understand the data. Graphs were also seen as useful but these needed careful explanation. The parents found Pareto fronts a particularly useful way of visualising the balance that might be struck between the differing demands of providing accessible services and centralising neonatal units to improve clinical outcomes.

The group also discussed the need to explain the methodology that produced the results. The parents did not feel that this needed to be done in great detail. However, they felt that the process behind the production of these results had to be made transparent to deal with concerns that units may be being closed merely to save money. Given the high numbers of potential scenarios generated by operational research, the parents found the evolutionary analogy helpful in explaining how these scenarios might be narrowed down, but care needed to be taken with language and terminology (e.g. referring to variations of a scenario as 'child' scenarios).

The parents felt that once a limited number of scenarios had been developed by operational research methods, they should be involved in any final decision-making about neonatal reorganisation. They felt that their local knowledge and experiential expertise would be crucial in making a fully informed final decision. Our work in this workshop demonstrated that it would be feasible and practical to do this.

Parents also felt that, when possible, the transfer of babies between neonatal units should be minimised. They reported that there are sometimes significant differences in the way care is delivered between units, which can create additional stress for families (e.g. because parents have to learn how a new unit works).

In this workshop, a proposed health economics interview schedule was also reviewed. The parents expressed concern about some of the terminology used and the order of questions. After the workshop, Sue Prosser facilitated revisions to the interview schedule, working closely with the qualitative researcher to reorder and rework the interview schedule with parental input. This included a practice/mock interview in a role play with Sue Prosser. The parents also reviewed the flyer that was used to attract volunteer interviewees. Copies of the advertising flyer and interview schedules used for the qualitative interviews with parents are provided in *Report Supplementary Material 2*.

April 2017 workshop

This was held in Exeter with parents in London able to join via Skype and was a final workshop to present the finding of our work to the parents and to discuss possible dissemination activities. The parents confirmed that they would potentially be interested in being involved in further dissemination work. We discussed what we had learnt and how parents might be involved in the decision-making about potential reorganisations of neonatal care. The parents were very clear that they felt that they could and should be involved in this decision-making process. They felt that the evidence for the clinical benefits of centralisation, including the weaknesses in this evidence, needed to be presented to parents in an unbiased way. They also felt that operational modelling potentially offered opportunities to make the decision-making process more open and transparent but that any final decisions should incorporate their local knowledge alongside the knowledge derived from operational research.

Parent involvement summary

In conclusion, we feel that our work has shown that it is feasible and practical to involve parents in operational research about service reconfigurations in neonatal care. It is important to do this to ensure that this research takes into account the issues and concerns of parents. Our work has also shown that parents can appreciate the complexities involved in making decisions about the reorganisation of neonatal services if they are provided with the relevant information in an accessible manner. Any such decision-making process is likely to benefit from this input.

Chapter 12 General discussion and conclusion

In this study, we have analysed and researched the organisation of neonatal services in England with a focus on demand and capacity issues. Specifically, we have used location analysis and simulation to investigate the geographic placement of services and capacity constraints. We have looked at the number of units required at different service levels, and the flow of patients in the system. In addition, we have examined the economic issues and modelled these against mortality outcomes, and we have engaged with parents to assess their preferences and priorities in relation to neonatal services.

In all of these areas, it is important at the outset to emphasise the variability and diversity of conditions that pertain across the English neonatal care system. For example, differences in geographic conditions and population densities (e.g. rural vs. metropolitan) play an important part in determining the effective placement of services. Size and level of unit also crucially have an impact on service organisation and hospitals often markedly differ in how they choose to organise their systems (e.g. in the deployment of staff and the interface with maternity services). Commissioning arrangements for neonatal care in the NHS also vary and this is likely to have an impact on how services are organised and reported. Turning to parental factors, accommodation and support varies markedly between units and the experience of parents will differ greatly in terms of financial, family and domestic circumstances, as well as geographic and physical needs. This leads to inevitable differences in the preferences and priorities of individual parents, although it should be noted that the overriding concern is the well-being of the mother and child.

Given the multiple dimensions of variability, it is challenging to develop generalised models that can be applied across all neonatal services in England and for all users and all providers. A flexible approach is required that can accommodate these differences yet still provide a framework for both understanding and guiding policy in order to provide the evidence base needed to develop new and improved models of care. Our research aims to provide a methodological framework that can begin to provide a basis to support evidence-informed decision-making in neonatal care across England.

We have included the more technical and detailed comments in relation to the specific components of the research in the relevant sections of the report. In the following sections, we summarise some of the salient points arising, list some key limitations, and outline key areas for further research in this field.

Location analysis

In approaching location analysis, we have focused on understanding the key aspects of capacity and demand in the system and how this relates to service organisation. For this, a comprehensive analysis of the data from across the units provides a necessary basis to understand the current state of neonatal service activity across England. Our analysis suggests that some aspects of the current system could be improved and we have modelled a series of 'what if' scenarios to assess the potential impact of different systems. Here, two approaches can be defined. First, it is possible to model idealised scenarios that provide optimised system outputs against the defined performance metrics. Although in most cases these hypothesised scenarios do not provide realistic or practical options, they do provide critical reference points in determining the upper limits to potential organisation. The hypothetical configurations also encourage policy-makers to 'think outside the box' in terms of considering options. The second approach is to start with the practical options for reorganisation (e.g. as suggested by policy-makers). These alternative scenarios can then be modelled and the likely effects of changes on key output metrics assessed using the methods outlined in this report.

It is inevitable that economies of scale exist, favouring the increased centralisation of services, especially for the more intensive levels of care. This policy imperative is further reinforced by the research findings that suggest that outcomes are improved in larger higher volume units.[151]

Simulation

Simulation, although still working with a simplified model of reality, allows for more complex behaviours to be explored than are possible in the genetic algorithm, which must explore hundreds of thousands of possible configurations. A simulation model may track infants through multiple levels of care and across multiple spells in different hospitals. Hospitals may also have restricted capacity in simulation modelling, forcing infants to be cared for in a unit located further away than their nearest appropriate unit, and units may be organised in networks in which infants are only cared for outside the home network when the network has no appropriate cots available.

The simulation modelling further added to our understanding of the behaviour of the system at a national level. Performance of the system was found to be very sensitive to capacity constraints, with significantly deteriorating performance from about 65% of capacity utilisation. This is attributable, first, to the relatively small size of neonatal units with little or no flexibility within a hospital to care for infant patients who cannot be cared for in the designated ward. Second, additional demand may come from infants who should be cared for at another hospital. The networked model of neonatal care allows for excessive demand to be absorbed by neighbouring units, but this movement of demand may reduce the ability of local units to meet their own local demand.

Higher-volume units were found to be more resilient to fluctuations in local demand because of the relatively smaller variation in demand in large units. This allows high-volume units to meet BAPM standards of care for local demand with fewer nurses. A more centralised model of care (with 90 neonatal units) was found to substantially meet BAPM standards of care with about 10% fewer nurses than the current configuration.

The simulation modelling confirmed results from the genetic algorithm location analysis: that configurations exist, at least theoretically, that can both increase the number of VLBW infants cared for in units that receive ≥ 100 VLBW infants per year, and improve access to care for parents. Modelling indicated that the number of NICUs was approximately right if all NICUs should have ≥ 100 VLBW infant admissions per year, but the locations of those NICUs are not currently optimal for achieving the joint aims of all NICUs receiving ≥ 100 VLBW infants per year and having NICUs as close to parents' home locations as possible.

The simulation tool developed may be applied to national configurations, as described in this report, but it may also be applied more locally.

Mortality

The EPICure study reported on outcomes for all births before 26 weeks in the UK and the Republic of Ireland for a period of 10 months in 1995.[152] A survival rate of 39% was found for the 20.2% of all infants who were admitted to a NICU. It has been reported that the survival rate for infants with 22^{+0} to 25^{+6} weeks of gestation who were admitted to a neonatal unit increased from 36% in 1994 to 47% in 2000–5[153] in a region of England. In the English data set for 2015/16 used here (which contained all types of neonatal unit), looking at the same gestational age group, 31.9% died, 58.7% exited neonatal care alive and 9.4% were still in neonatal care when the data were reported. Although the methodology is not identical, this would suggest that survival rates have continued to improve.

Our estimates for infant mortality are for those born in a high-volume hospital, which includes the benefits of antenatal care in the hospital of birth as well as the postnatal care in the NICU. A very preterm infant born in a high-volume hospital (> 100 infants weighing < 1500 g per year) has a 4.5% lower risk of death in this unit than in other hospitals. Slightly larger effects were observed when using distance to a high-volume hospital as the instrument. An infant born at < 32 weeks' gestation being born in a hospital with a tertiary NICU unit does not appear to result in any difference in terms of the risk of death compared with other hospitals.

Costs

Our economic analysis has concentrated on the underlying neonatal costs for both the NHS and parents in relation to service organisation. For the NHS, costs need to be clearly differentiated from prices, given the variance in contractual and commissioning arrangements for neonatal services in England. This report does not address issues of price and commissioning that fall outside the scope of the study.

In relation to NHS neonatal costs, there is a clear need to go beyond the arguably crude models that are currently used. The site visits and discussion with policy-makers made it clear that the neonatal HRG reference costs currently do not accurately reflect the real overheads of neonatal care, mainly because units typically average neonatal nursing costs across all infants and do not use the BAPM guidelines[2] to attribute nursing to the different levels of care. Therefore, in our evaluation section (see *Chapter 9*) we ran two sets of analyses to (1) calculate the cost-effectiveness of high-volume hospitals compared with other hospitals and (2) cost three simulations from the computer model. The first method used the BAPM 2001 reference costs, which matched the payment system used in 2013–15 that was used to reimburse units, and so was the opportunity cost to hospitals. The second method calculated the costs of the simulations based on the predicted staffing predictions from simulations.

As of 2018, data are available using the 2016 HRG codes that more accurately reflect BAPM guidance for nursing. In addition, more is now known of the thresholds in consultant time and a detailed health economic analysis of these costs, to develop cost models based on these data sets, would more accurately capture economies of scale and scope. In small units, for instance, there may be granularity constraints such that provision of a specific level of care requires at least one specialist nurse even though that nurse may not be fully utilised.

The BLISS data set gave a snapshot of costs for neonatal care for those mothers who are part of the BLISS network, but there were a lot of incomplete data and the sample was not randomly selected, so caution is needed regarding the results. Incomplete data may have arisen because the survey included many questions, and there were problems with coding some of the open questions. In future studies, we would recommend much shorter surveys with simple click boxes to improve completion rates, focusing on only a few key issues. A discussion with PPI members about whether or not these costs would change with different configurations suggests that food costs are unaffected by service configurations, but costs of childcare, travel and overnight stays could be affected.

Factors that families and policy-makers would like to see taken into consideration in determining service configuration

The qualitative study with families suggested that families felt that the following aspects were important to their experience of neonatal care: the baby's and mother's health, communication by medical teams and support for families. The interviews with policy-makers also raised the issues of the baby's health and mother's well-being, as well as the need to staff units appropriately and recruit and retain staff.

The qualitative study with parents raised questions about the ability and willingness of parents to trade off health attributes (the baby's and mother's health) with the process attributes (communication by medical teams and support for families). Although no trade-off questions were asked in the patient interviews, it became clear that mothers were unlikely to want to sacrifice these 'core' aspects of their health and their baby's health for improvements in process outcomes. This raises questions about the use of a DCE with combined health and non-health outcomes, as parents would always choose the configuration that favoured the best health for the infant and the mother. However, if a reconfiguration maintained health but affected process outcomes, then trade-offs and a DCE become more feasible.

Study limitations

There are a number of key limitations and constraints to our study (many of which are outlined above). Some of the most significant limitations are:

- Our analysis is clearly hampered by the difficulties inherent in assessing morbidity (as distinct from mortality outcomes) in neonatal care.
- In this project, we have looked at location optimisation algorithms for both childbirth and neonatal care. The planning of each of these in the NHS in England is the responsibility of separate organisations: maternity and childbirth planning is the responsibility of Clinical Commissioning Groups, whereas neonatal care planning is the responsibility of NHS England Specialised Commissioning.
- Our study assumes that patients attend their closest appropriate unit. When the travel times to units are similar, factors other than travel time may dominate the choice of unit.
- Our study is based on road travel times. The use of public transport may, at times, change the preferred hospital, and is also likely to change the absolute travel times to all hospitals.
- In this study, we have not focused on the location of the phase of any surgical care that must be carried out in specialist neonatal surgical units. It is likely that specialist surgical care would usually be carried out in NICUs.
- It is not clear whether the advantages in outcome associated with high-volume NICUs are attributable solely to the NICU, or are also because those units will often tend to be associated with large maternity units (with the associated outcome advantages of more consistent obstetric consultancy presence). It is not clear how much clinical advantage will be gained by centralising only neonatal intensive care, centralising only childbirth care or by combining both. Joint planning of maternity and neonatal care would help provide hubs of clinical excellence with both ≥ 6000 births per year and NICUs admitting ≥ 100 VLBW infants per year.
- A limitation of the mortality estimates is that they measure the effectiveness of those infants born in high-volume hospitals or NICUs, but not the effectiveness of postnatal transfers ($\approx 20\%$ of those with a gestational age of < 33 weeks). In addition, the effectiveness of the in utero transfer is likely to be dependent on the point at which antenatal cover began. For example, hospitals may be more effective at reducing mortality for earlier in utero transfers (which tend to be the highest risk transfers) because of the improved antenatal cover.

Opportunities for further work

In terms of extending this research and developing its full potential to inform policy, much still needs to be achieved and we would point to the following specific areas for further research to build on this evidence base:

- There is a need to model the interface between maternity and neonatal services to fully understand the dynamics of the system and its impact on outcomes and costs. It may be desirable to model births and NICU admissions together to identify the best locations of hospitals to combine the Royal College of Obstetricians and Gynaecologists recommendation[22] of ≥ 6000 births for each obstetric unit where possible and the BAPM recommendation[21] that all NICUs receive ≥ 100 VLBW infants per year.
- The current study combined geographic location analysis with simulation modelling for neonatal care, allowing for an investigation of the impact of altered configuration on the number of neonatal nurses required to cope with variation in workload. It is possible to do the same for childbirth, combining a geographic location model with a simulation of labour-ward workload (the latter has previously been carried out in isolation by the authors[154]).
- Methods should be developed to assess infant morbidity (as well as mortality) to fully analyse outcomes and the impact on cost-effectiveness of treatment and service delivery options.
- The effectiveness and impact of transfers between hospitals should be explored (e.g. early and late in utero transfers and postnatal transfers), drawing on data available for the antenatal and postnatal periods.

- The extent to which high-volume hospitals affect short-term and long-term morbidities, taking account of both the antenatal period and the postnatal period, should be considered.
- There is a need to understand more fully the effect of network boundaries on the organisation of services.
- There is a need to explore and evaluate in more detail how effective visualisation tools can be deployed across a range of contexts and for a range of stakeholders to convey the key findings in this area.
- A DCE should be fully implemented with parents of neonatal infants based on the initial preparatory work presented in this study to systematically assess parental preferences in neonatal care.

Dissemination and outputs

Publications

A range of publications and presentations are planned on the basis of this work. These develop the themes and present them in more technical detail or explore some of the key methodological issues. These publications and presentations comprise:

- a detailed analysis of the geographic analysis applied to maternity units in the context of current guidance from the Royal College of Obstetricians and Gynaecologists
- more detailed and technical approaches to the mathematical methods used for the location analysis in this study
- an expansion of the technical aspects of the health economic modelling and a comparison of alternative approaches to cost and mortality modelling
- an expanded examination of the use of visualisation tools to communicate research output based in a thorough user-centred requirements analysis
- a paper that investigates the differences between PPI workshops and qualitative research approaches, using neonatal care and this study as a case study example.

Tools

The methodological framework presented in this report, we believe, provides the basis for a tool set that could be used to support an evidence base for policy-makers in neonatal health and care. Although considerable further development would be required to fully implement this framework, we believe that it has the potential to be readily adapted for general use.

Conclusion

In many respects, this study has generated at least as many questions as answers. However, it has demonstrated a methodological approach and framework for the in-depth analysis of key issues in organising neonatal care for the NHS in England. In particular, we would point to our detailed location analysis for neonatal units in England as an important contribution to a more systematic and evidence-based approach to planned service configuration in the context of the evidence for improved outcomes in high-volume NICUs. In addition, the health economic analysis and mortality modelling point to key issues for differing configurations of care gives key insights into the cost implications for policy alternatives. Our parental interviews and qualitative research provide the crucial perspective of parents and highlighted a number of their key issues and concerns. Likewise, the interviews with policy-makers in neonatal care provided the critical context in which decision-making and consideration of service options take place.

It is clear that much more work needs to be undertaken to develop a comprehensive approach to the challenges facing the NHS for co-ordinated childbirth and neonatal care in England. However, we believe that this study significantly advances this cause and points to a range of further research needs in this context.

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Contribution of authors

Emma Villeneuve (Associate Research Fellow) developed the location analysis and mathematical modelling, contributed to obtaining permissions from all neonatal units to source NDAU data. As the first author, she co-ordinated and managed the writing of the report as well as writing relevant sections. Area of specialty: mathematical modelling and location analysis.

Paolo Landa (Associate Research Fellow) conducted the literature review, analysed family costs, prepared data and helped develop results for mortality and costs modelling. He conducted policy-maker interviews along with Anne Spencer and undertook the reading and coding of parent interviews and the development of the NVivo framework with Katie Kelsey. Area of specialty: health economic analysis and modelling.

Michael Allen (Honorary Research Fellow, Operational Research) framed the location analysis and simulation work, performed the descriptive analysis of data and the simulation work, and wrote part of the report. Area of specialty: mathematical modelling and simulation modelling.

Anne Spencer (Associate Professor, Health Economics) developed the framework for health economics analysis and conducted site visits and policy interviews along with Paolo Landa. She also wrote the initial ethics application for the qualitative study and wrote part of the report. Area of specialty: health economics.

Sue Prosser (Senior Neonatal Nurse) advised on neonatal unit working practices and assisted in parent involvement. She co-led recruitment, facilitated the PPI group and supported the qualitative researcher and health economics team. She also contributed to the PPI section of the report. Area of specialty: neonatal nursing.

Andrew Gibson (Associate Professor, PPI) co-led, with Sue Prosser, the PPI research work in the project and co-wrote the PPI section of the report. Area of specialty: PPI.

Katie Kelsey (Qualitative Researcher) conducted parent interviews, carried out qualitative analysis (developing the coding framework with Paolo Landa, coding with NVivo and analysis of responses using framework matrices) and wrote part of the report.

Ruben Mujica-Mota (Senior Lecturer in Health Economics) conducted and developed the mortality models and the cost modelling on LOS. Area of specialty: quantitative research.

Brad Manktelow (Associate Professor, Statistics) contributed to the design of the study and the interpretation of the data and reviewed the report as a whole. Area of specialty: neonatal modelling and mortality analysis.

Neena Modi (Professor, Neonatal Medicine) advised on clinical aspects, oversaw the acquisition of data from the NNRD and reviewed the final report. Area of specialty: neonatal care services.

Steve Thornton (Professor, Obstetrics and Gynaecology) provided clinical guidance for research relating to maternity services and reviewed the report, providing input to relevant sections. Area of specialty: obstetric care.

Martin Pitt (Associate Professor, Health-care Modelling) was the chief investigator for the research and oversaw the management and co-ordination of the project as a whole. He reviewed, amended and finalised the report and authored some sections (e.g. data visualisation). Area of specialty: modelling and simulation applied to health care.

Data-sharing statement

All data requests should be submitted to the corresponding author for consideration. Access to anonymised data may be granted following review.

Patient data

This work uses data provided by patients and collected by the NHS as part of their care and support. Using patient data is vital to improve health and care for everyone. There is huge potential to make better use of information from people's patient records, to understand more about disease, develop new treatments, monitor safety, and plan NHS services. Patient data should be kept safe and secure, to protect everyone's privacy, and it's important that there are safeguards to make sure that it is stored and used responsibly. Everyone should be able to find out about how patient data are used. #datasaveslives You can find out more about the background to this citation here: https://understandingpatientdata.org.uk/data-citation.

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Appendix 1 Location analysis

Villeneuve charts for resilience probability

Definition of the resilience probability

In order to analyse the possible scenarios, it is interesting to highlight which locations are more likely to appear in the optimal configurations (i.e. those configurations that make up the Pareto front). To do so, we compute the resilience probability of every location in accordance with the following method:

1. Select the Pareto front configurations with 35 to 55 units (current number 45 ± 10). This allows us to select configurations close to the current state.
2. Select the Pareto front configurations in the highest quartile of the proportion of patients both attending units with ≥ 100 VLBW infants per year and living within 30 minutes from the closest unit. This allows us to select the best-performing configurations.
3. Compute the probability $p(u_i|h)$ of each unit u_i to appear in the selected '*h*-configurations' (configurations with *h* units):

$$p(u_i|h) = \frac{Number\ of\ h\text{-}configurations\ with\ u_i}{h \times number\ of\ h\text{-}configurations}. \quad (7)$$

Such probability verifies:

$$\forall h \sum_{i=1}^{H} p(u_i|h) = 1. \quad (8)$$

1. Compute the probability $p(u_i)$ of each unit u_i to appear in the selected configurations:

$$p(u_i) = \frac{1}{(h_{max} - h_{min} + 1)} \sum_{h=h_{min}}^{h_{max}} p(u_i|h). \quad (9)$$

Such probability verifies:

$$\sum_{i=1}^{H} p(u_i) = 1. \quad (10)$$

The locations that appear the most often in the optimal configurations will have a higher resilience probability. Such an indicator is useful in order to select the locations that are the most resilient to network changes.

Reading a Villeneuve resilience chart

These charts display the resilience or optimality probability of the locations of four ODNs, as well as their existing levels of care.

The Villeneuve resilience chart provides a means to assess the relative probability that a neonatal unit location appears in an optimal configuration (as output by the genetic algorithm used for location analysis) and is therefore likely to be a good location to place a NICU. In *Figure 20*, unit locations in four separate networks are shown (represented by the upper level of bars). For each location, the probability of each existing unit appearing in an optimal configuration (coloured bar) as well as the unit's existing care level [NICU, LNU or SCU (lower level of bars)] are shown.

In the example in *Figure 20*, it can be seen that some lower level units (e.g. Dartford and Haywards Heath) are very likely to be in favourable locations for the establishment of a NICU.

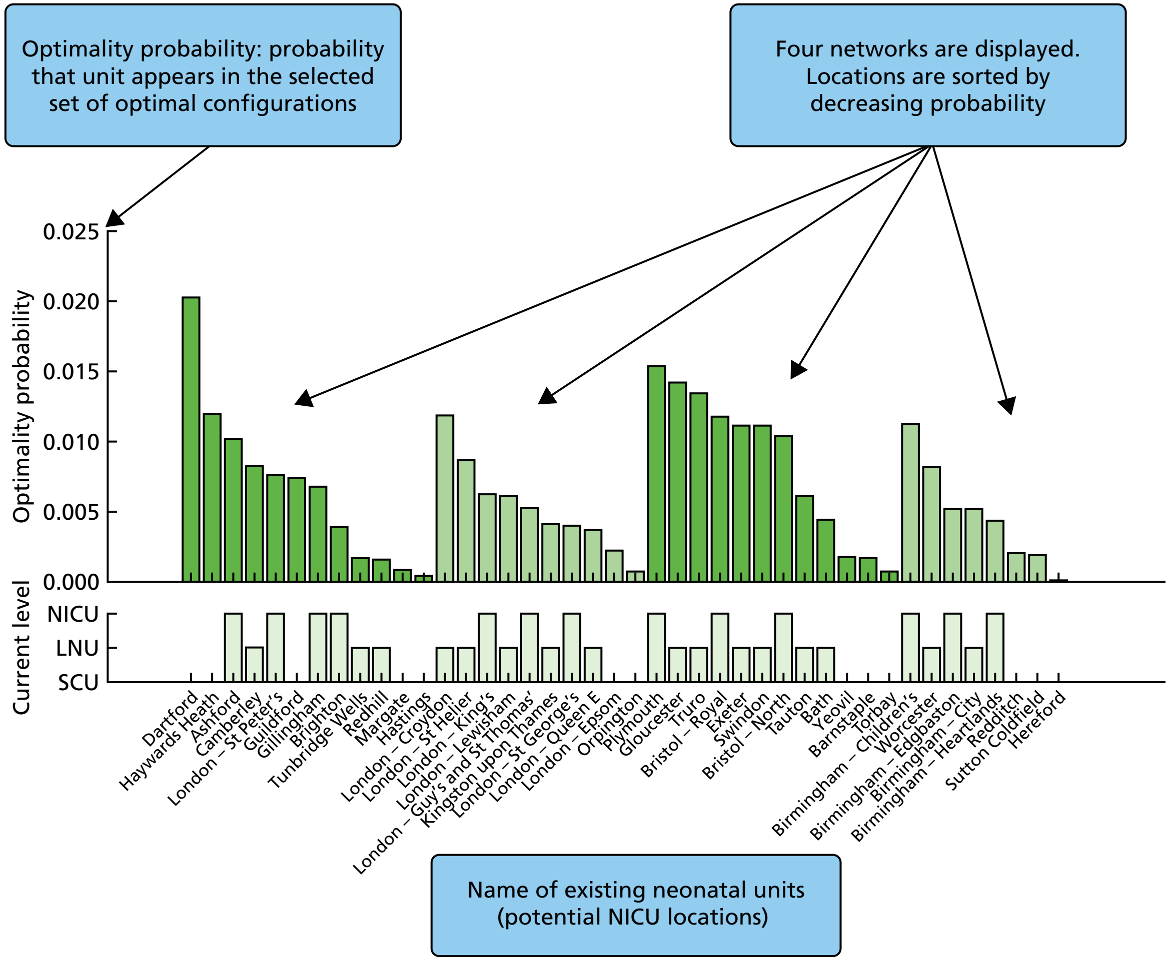


FIGURE 20 Elements within a Villeneuve chart.

DOI: 10.3310/hsdr06350

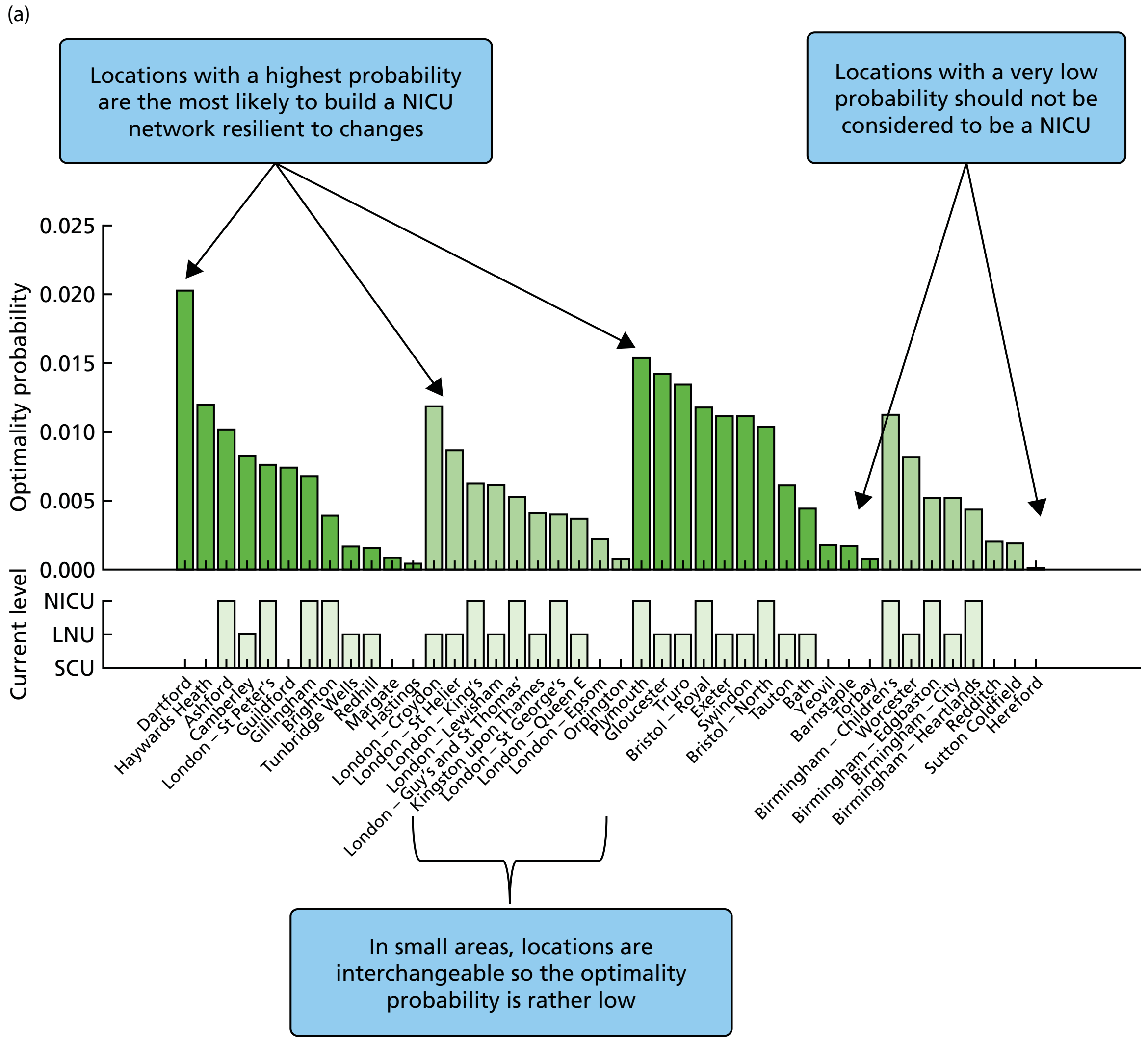


FIGURE 21 Annotated elements of Villeneuve charts. (*continued*)

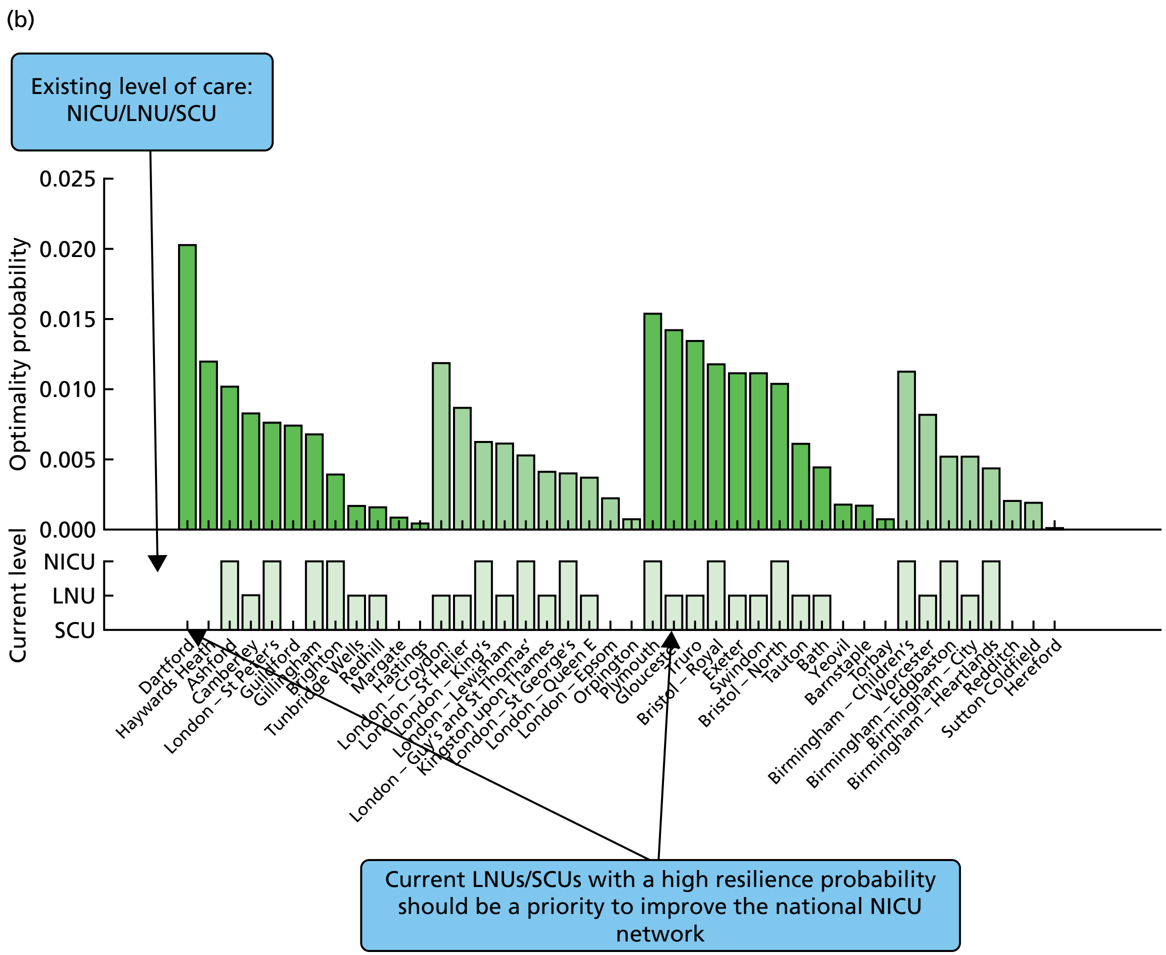


FIGURE 21 Annotated elements of Villeneuve charts.

Resilience charts for other networks

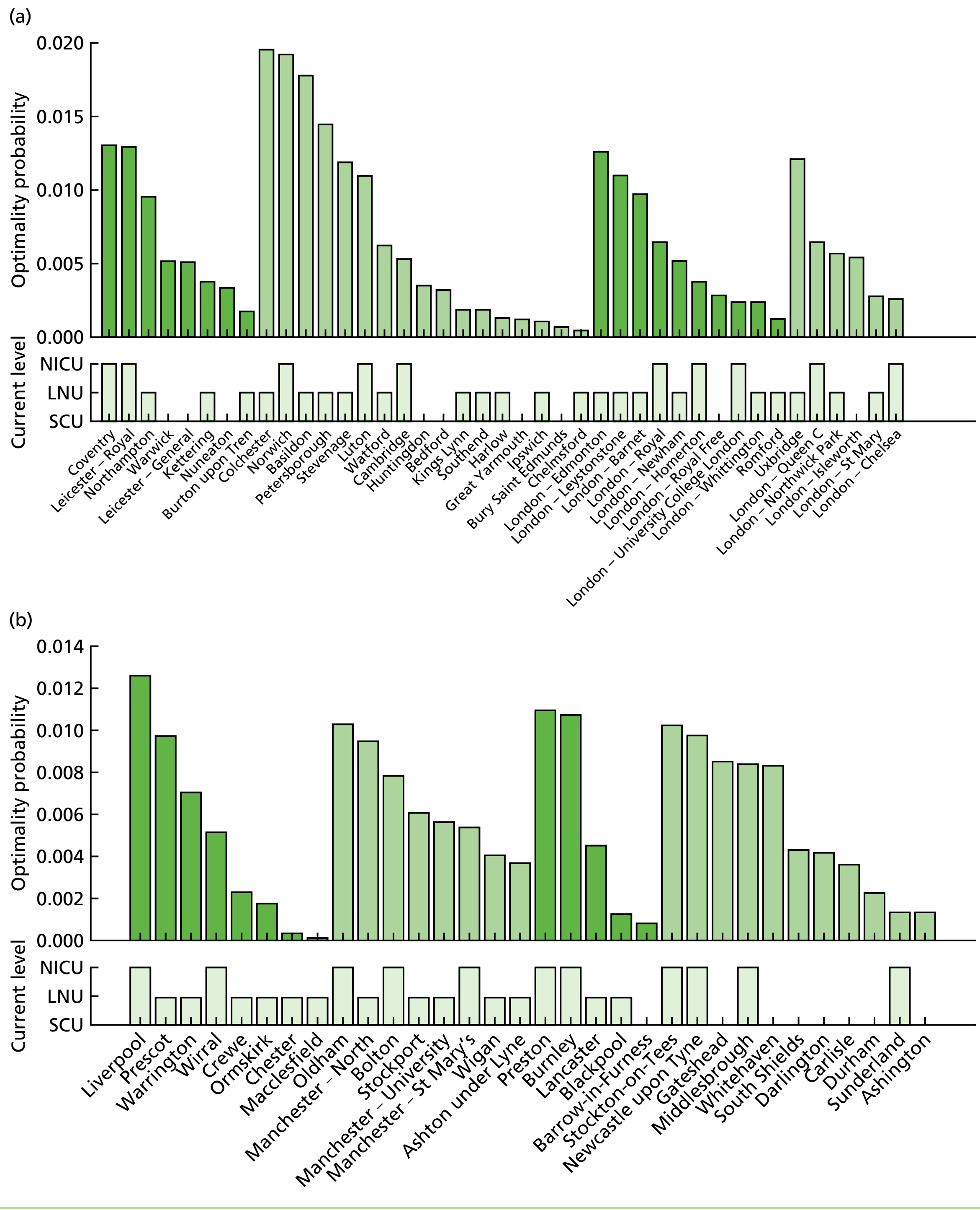


FIGURE 22 Villeneuve charts for specific neonatal network areas. (*continued*)

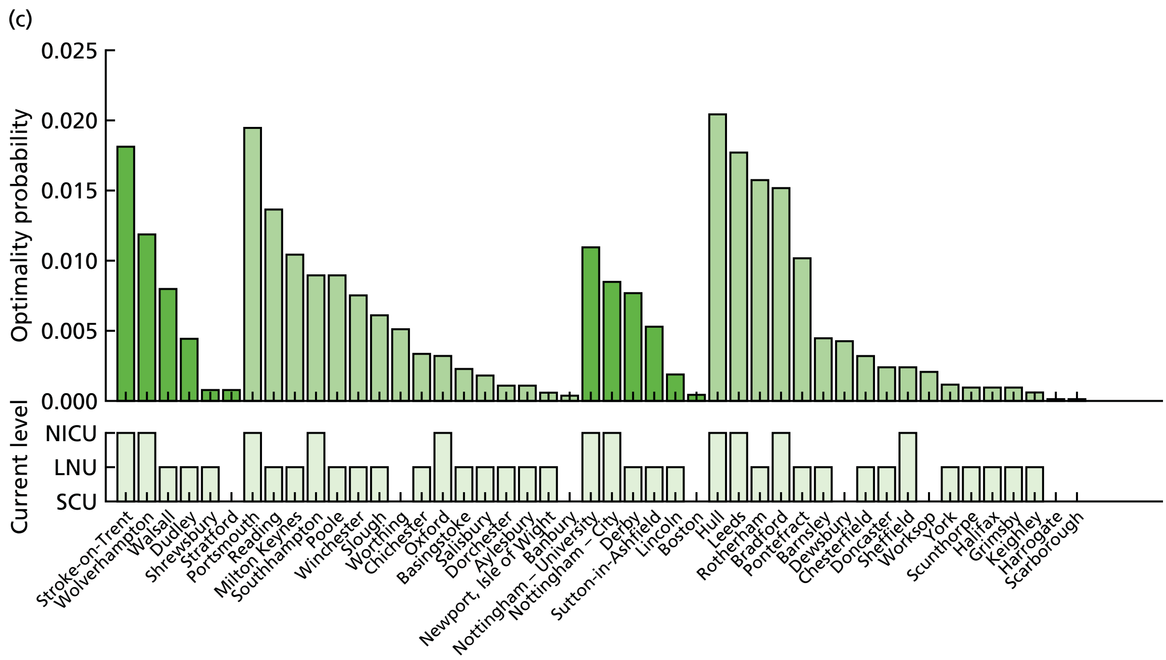


FIGURE 22 Villeneuve charts for specific neonatal network areas.

Definition of the resilience scenario score

The optimisation process provides thousands of Pareto front scenarios. To select a representative scenario $\boldsymbol{u} = \{u_1, \ldots, u_h\}$, we can compute the resilience score $R(u)$ defined as follows for all scenarios:

$$R(u) = \sum_{i=1}^{h} p(u_i). \tag{11}$$

Access to intensive care for very low-birthweight infants

(a)

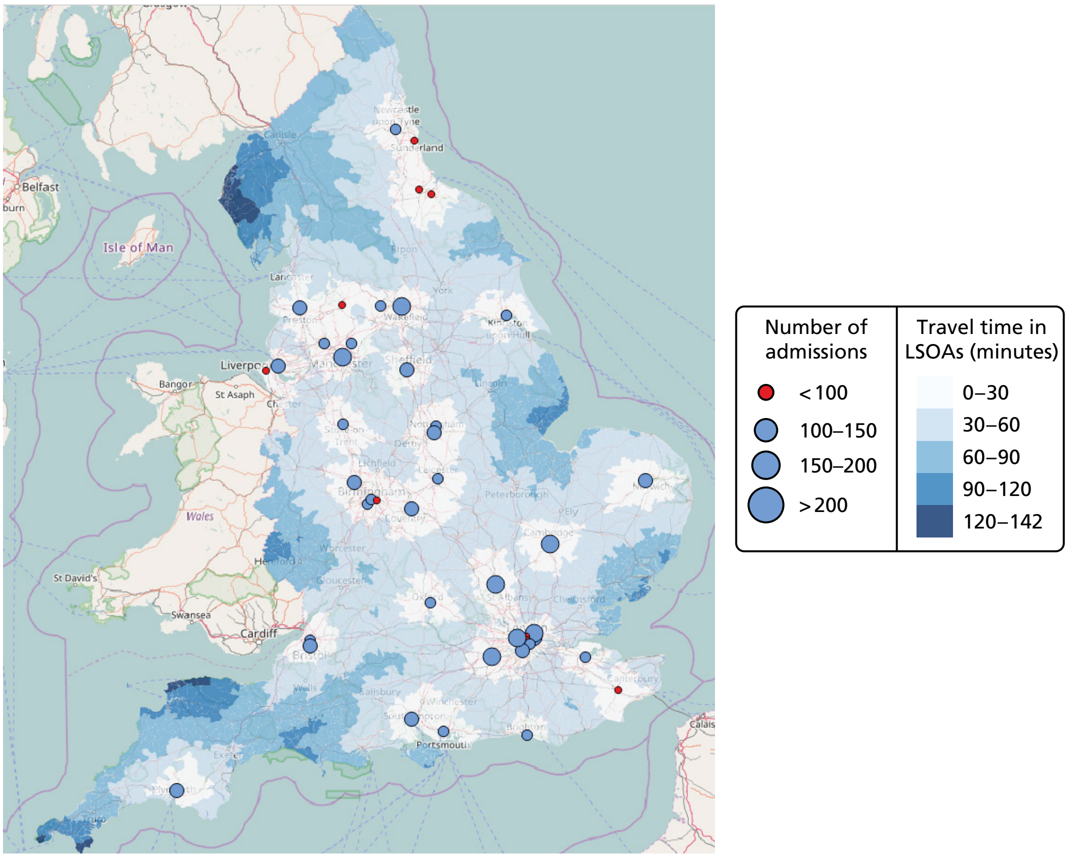
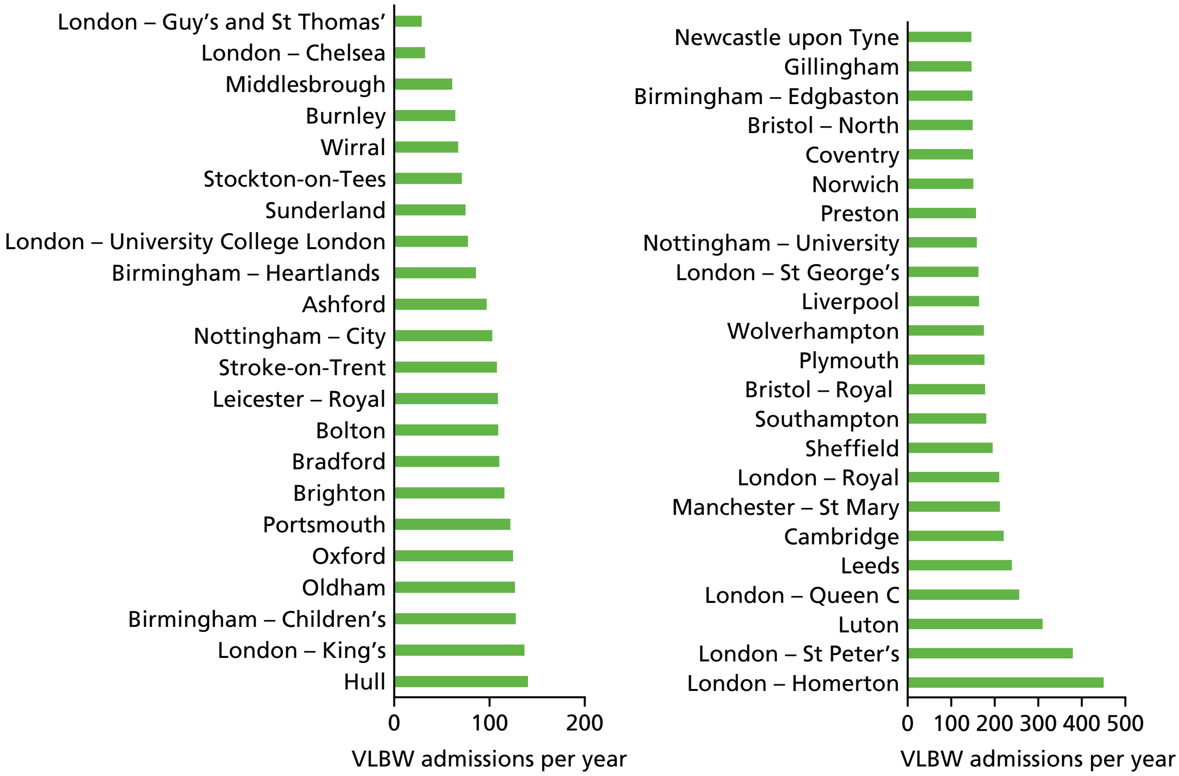


FIGURE 23 Current configuration of NICUs in England with 45 NICUs. (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); (b) VLBW admissions per year; and (c) key performance indicators. (*continued*)

(b)

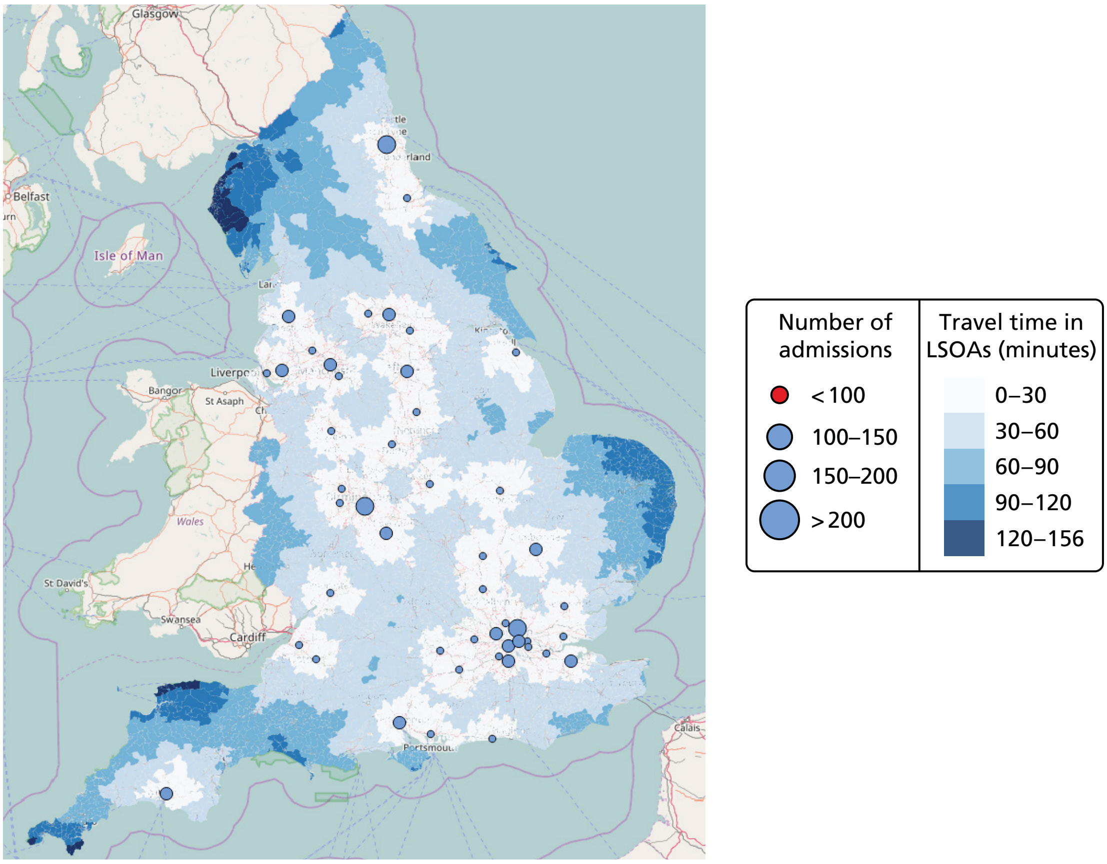


(c)

Average travel time	28 minutes
Maximum travel time	142 minutes
Mothers within 30 minutes of closest NICU	65%
VLBW infants attending unit with ≥ 100 VLBW infants per year	90%
Minimum number of VLBW admissions	29
Maximum number of VLBW admissions	450
Mothers and VLBW infants within 30 minutes of closest NICU and attending a unit with ≥ 100 VLBW infants per year	57%

FIGURE 23 Current configuration of NICUs in England with 45 NICUs. (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); (b) VLBW admissions per year; and (c) key performance indicators.

(a)



(b)

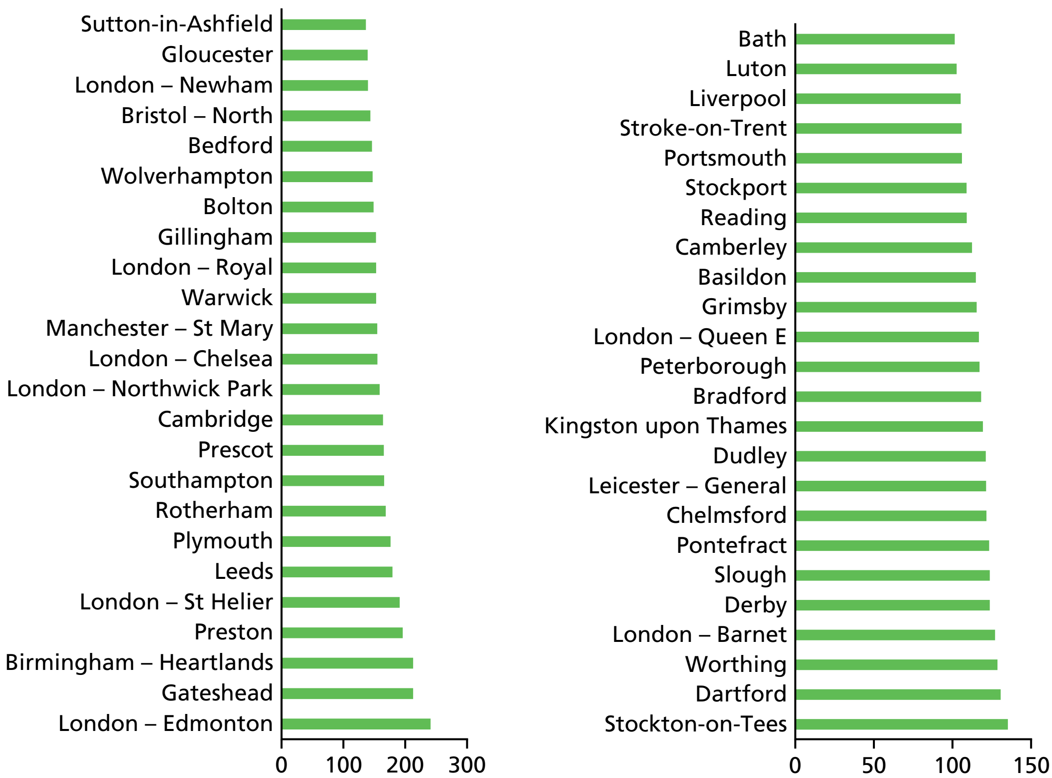


FIGURE 24 Example configuration of NICUs in England (48 NICUs). (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); and (b) VLBW admissions per year; and (c) key performance indicators. (*continued*)

(c)

Average travel time	26 minutes
Maximum travel time	143 minutes
Mothers within 30 minutes of closest NICU	73%
VLBW infants attending unit with ≥ 100 VLBW infants per year	100%
Minimum number of VLBW admissions	101
Maximum number of VLBW admissions	241
Mothers and VLBW infants within 30 minutes of closest NICU and attending a unit with ≥ 100 VLBW infants per year	73%

FIGURE 24 Example configuration of NICUs in England (48 NICUs). (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); and (b) VLBW admissions per year; and (c) key performance indicators.

(a)

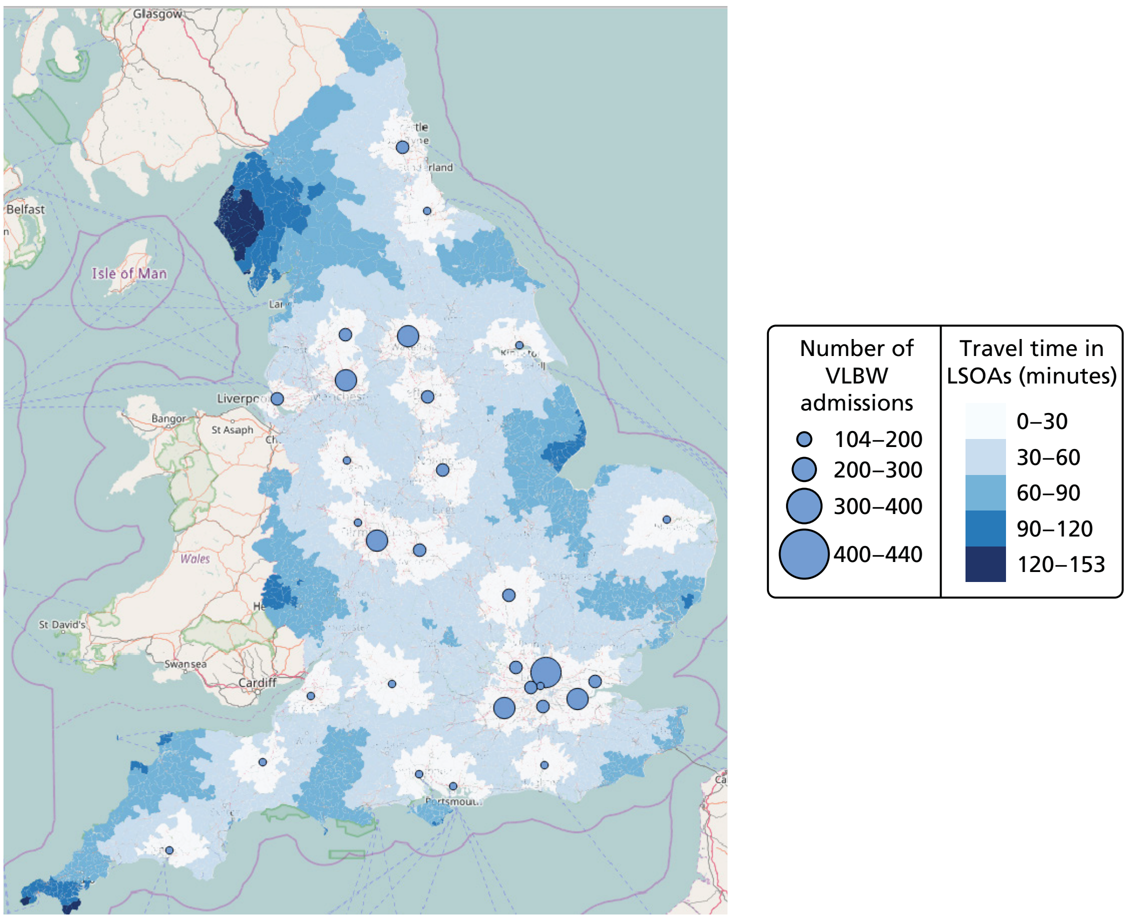
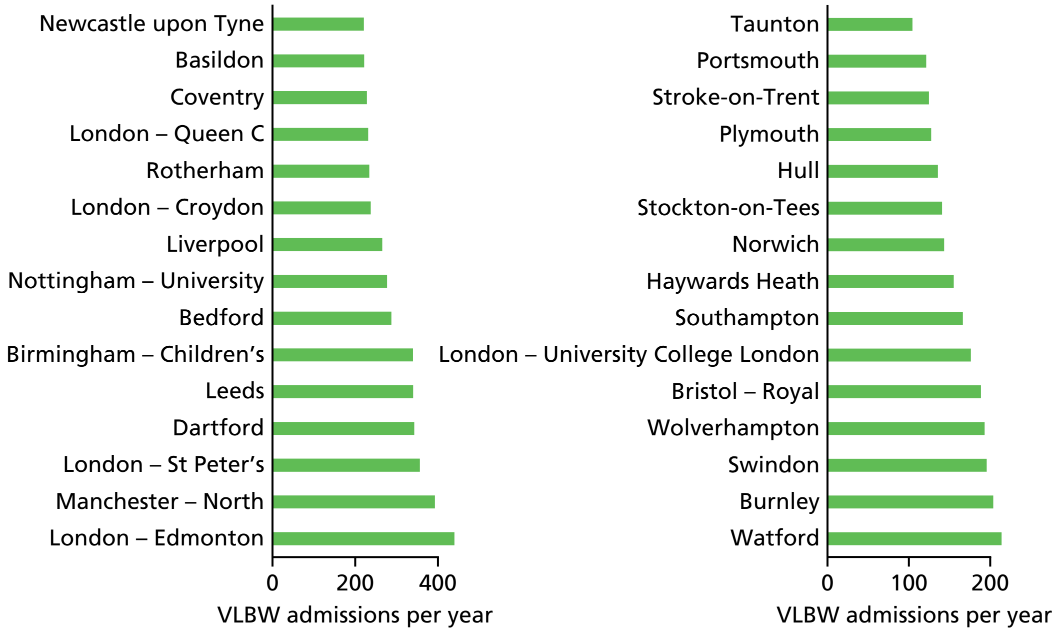


FIGURE 25 Example configuration of NICUs in England (30 NICUs). (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); and (b) VLBW admissions per year; and (c) key performance indicators. (*continued*)

(b)



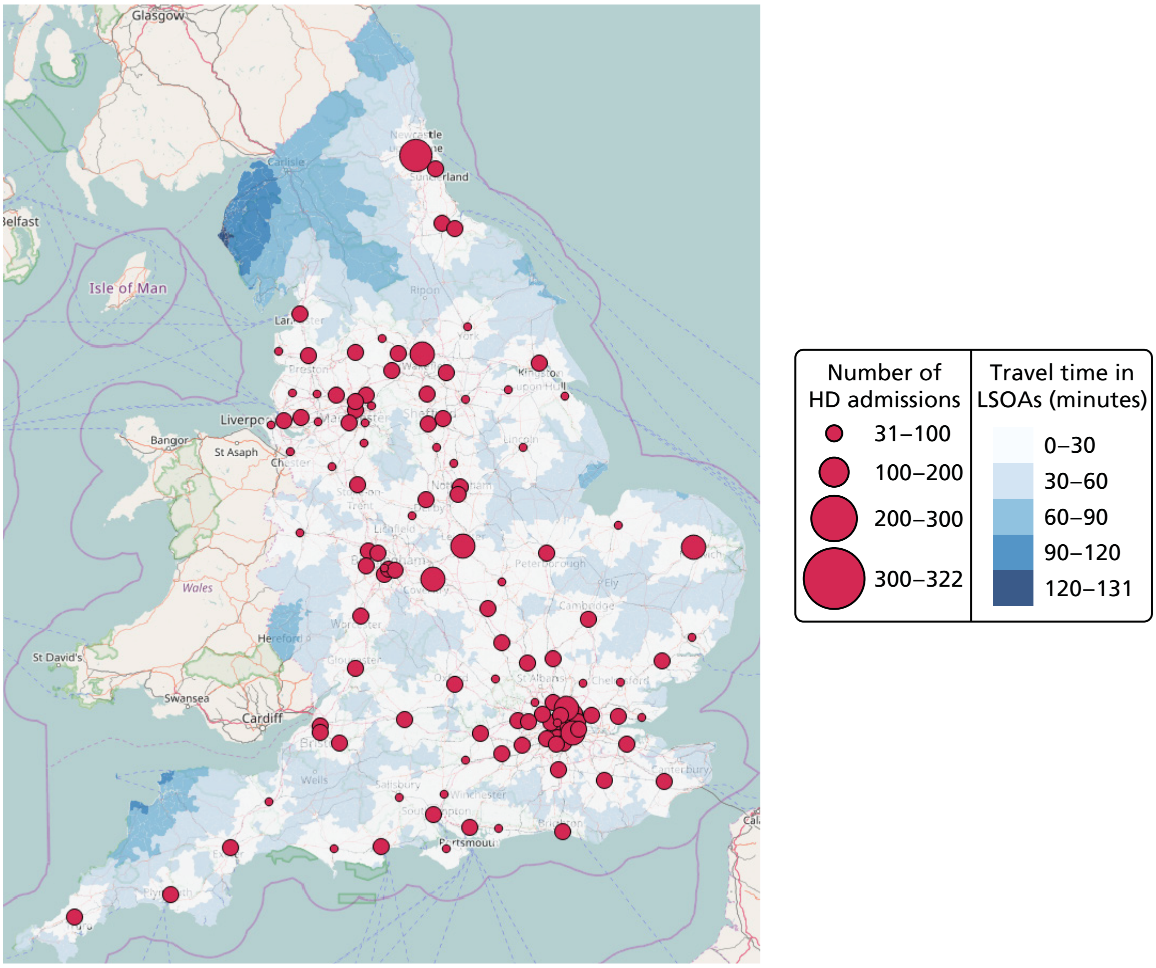
(c)

Average travel time	29 minutes
Maximum travel time	153 minutes
Mothers within 30 minutes of closest NICU	64%
VLBW infants attending unit with ≥ 100 VLBW infants per year	100%
Minimum number of VLBW admissions	104
Maximum number of VLBW admissions	440
Mothers and VLBW infants within 30 minutes of closest NICU and attending a unit with ≥ 100 VLBW infants per year	64%

FIGURE 25 Example configuration of NICUs in England (30 NICUs). (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); and (b) VLBW admissions per year; and (c) key performance indicators.

Access to high-dependency care

(a)



(b)

Average travel time	17 minutes
Maximum travel time	131 minutes
Mothers within 30 minutes of closest NICU	90%
Minimum number of HD admissions	31
Maximum number of HD admissions	322

FIGURE 26 Configuration with current 45 NICUs and current 78 LNUs with high-dependency demand (*Tables 22a* and *b*). HD, high dependency. (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); and (b) key performance indicators.

TABLE 22a Current NICUs relating to *Figure 26*

Existing NICUs (n = 45)	High-dependency demand
Newcastle upon Tyne	321.6
Leeds	287.7
Coventry	248.4
Norwich	203.6
Leicester – Royal	202.4
Gillingham	197.1
Birmingham – Heartlands	193.9
Portsmouth	187.0
Ashford	181.7
Birmingham – Children's	177.1
London – Queen C	173.8
Bradford	172.5
Sunderland	169.4
Brighton	169.4
Bolton	159.5
Stroke-on-Trent	158.5
Bristol – Royal	157.6
Stockton-on-Tees	157.5
Luton	151.1
Oxford	151.0
London – St Peter	150.0
Hull	149.3
Cambridge	148.3
Manchester – St Mary	147.9
London – King's	142.3
Nottingham – University	138.9
Liverpool	137.1
Burnley	136.5
Oldham	129.8
Birmingham – Edgbaston	128.1
Middlesbrough	120.8
Southampton	117.3
Plymouth	116.7
London – Royal	114.9
London – Homerton	114.6

continued

TABLE 22a Current NICUs relating to *Figure 26* (*continued*)

Existing NICUs (*n* = 45)	High-dependency demand
Nottingham – City	113.0
Preston	112.6
Bristol – North	108.3
London – St George	107.9
Sheffield	107.5
Wolverhampton	106.7
Wirral	88.7
London – Guy's and St Thomas'	65.4
London – Chelsea	62.6
London – University College London	57.7

TABLE 22b Current LNUs relating to *Figure 26*

Existing LNUs (*n* = 78)	High-dependency demand
London – Edmonton	215.0
London – Lewisham	208.3
London – Northwick Park	189.3
London – Newham	188.0
Uxbridge	183.3
London – Leystonstone	178.1
Basildon	175.6
London – Croydon	172.1
Exeter	170.2
Peterborough	164.5
Prescot	164.0
Milton Keynes	162.1
Walsall	159.6
Gloucester	157.1
Kingston upon Thames	153.7
Romford	150.6
London – Queen E	148.8
Camberley	145.4
Dudley	143.6
Rotherham	142.0

TABLE 22b Current LNUs relating to *Figure 26* (*continued*)

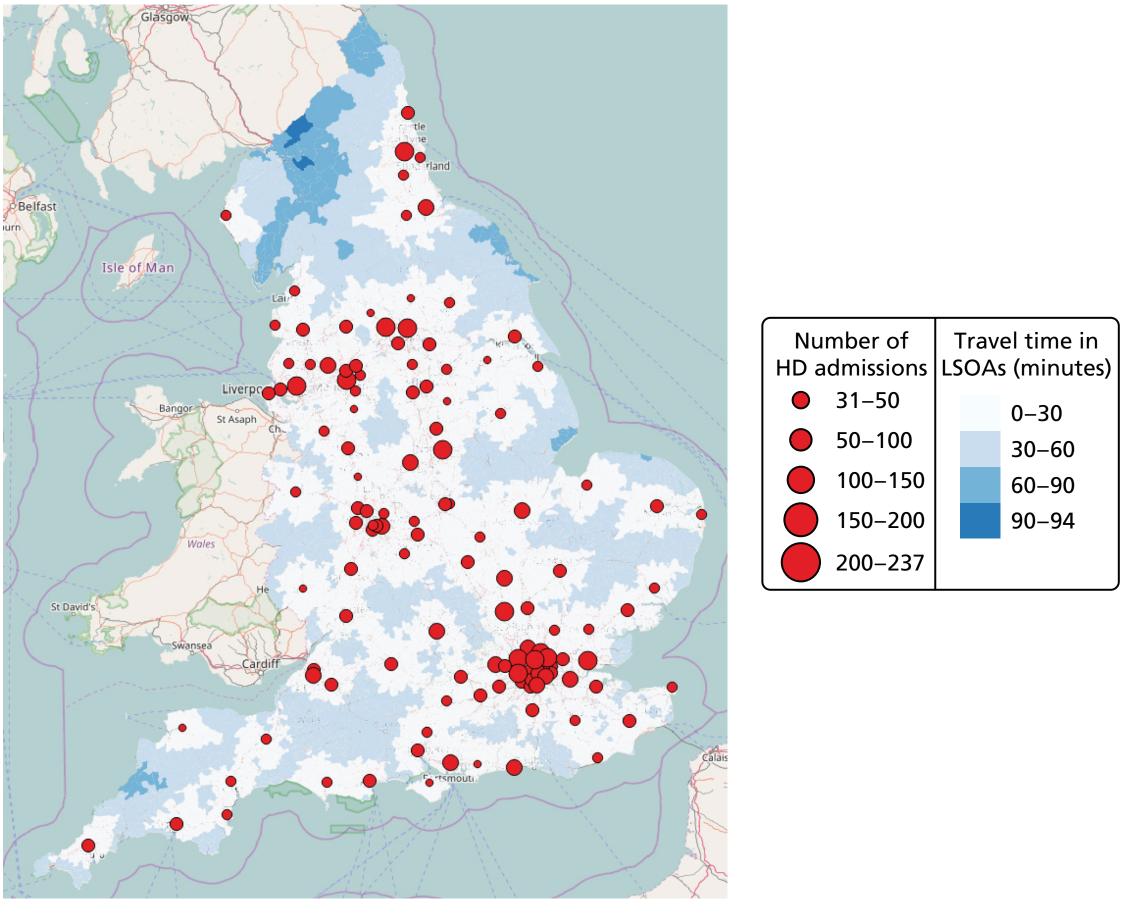
Existing LNUs (*n* = 78)	High-dependency demand
Reading	140.2
Tunbridge Wells	138.7
Manchester – North	135.4
Redhill	132.1
Slough	130.3
London – Whittington	129.3
London – Barnet	129.3
Worcester	129.2
Swindon	124.9
Lancaster	123.8
Northampton	122.2
Halifax	121.1
Bath	120.6
Derby	119.8
Poole	119.7
Barnsley	116.1
Stevenage	115.8
London – St Helier	108.8
Colchester	108.0
Manchester – University	106.7
Truro	105.0
Pontefract	103.7
York	98.8
Shrewsbury	97.5
Ipswich	95.7
Sutton-in-Ashfield	93.4
Doncaster	91.5
Harlow	89.2
Watford	88.0
Chichester	87.4
Taunton	87.2
Ashton under Lyne	84.2
Wigan	84.1

continued

TABLE 22b Current LNUs relating to *Figure 26* (*continued*)

Existing LNUs (n = 78)	High-dependency demand
Warrington	83.7
Kettering	81.4
Birmingham – City	81.1
Lincoln	79.9
Basingstoke	78.7
Chelmsford	77.7
Stockport	75.7
Aylesbury	75.4
Blackpool	72.5
Grimsby	72.2
King's Lynn	68.3
Southend	68.1
Crewe	67.4
Dorchester	63.9
Chesterfield	62.9
Ormskirk	60.0
Burton upon Trent	59.7
Salisbury	58.0
London – St Mary	57.7
Winchester	51.6
Chester	50.8
Scunthorpe	45.7
Keighley	43.4
Macclesfield	40.1
Newport, Isle of Wight	30.7

(a)



(b)

Average travel time	16 minutes
Maximum travel time	94 minutes
Mothers within 30 minutes of closest NICU	92%
Minimum number of HD admissions	31
Maximum number of HD admissions	237

FIGURE 27 Example of optimal configuration with 48 NICUs and 78 LNUs with high-dependency demand (*Tables 23a* and *b*). HD, high dependency. (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); and (b) key performance indicators.

TABLE 23a Configuration of NICUs relating to *Figure 27*

NICUs (n = 48)	High-dependency demand
London – Northwick Park	237.3
Manchester – St Mary	234.7
Prescot	232.7
London – Edmonton	223.6
Leeds	216.4
Basildon	213.5
Bradford	211.1
Gateshead	203.3
Luton	200.2
Birmingham – Heartlands	191.8
London – Newham	187.8
Portsmouth	187.0
Derby	184.1
Stockton-on-Tees	184.1
London – Royal	183.8
London – Barnet	182.9
Worthing	180.4
Dartford	179.1
London – Chelsea	169.7
Peterborough	169.3
Bolton	167.6
Bedford	153.4
Slough	152.0
Gillingham	148.4
Camberley	145.3
Dudley	143.6
Cambridge	143.0
Stroke-on-Trent	142.0
Reading	140.2
Liverpool	137.1
Gloucester	135.7
Sutton-in-Ashfield	131.6
Southampton	128.9
Bath	127.2

TABLE 23a Configuration of NICUs relating to *Figure 27* (*continued*)

NICUs (*n* = 48)	High-dependency demand
Rotherham	124.6
Kingston upon Thames	120.8
Preston	112.6
Plymouth	111.7
London – St Helier	108.8
Bristol – North	108.3
Wolverhampton	104.3
London – Queen E	102.3
Pontefract	100.2
Warwick	98.6
Stockport	85.4
Chelmsford	77.7
Grimsby	72.2
Leicester – General	67.3

TABLE 23b Configuration of LNUs relating to *Figure 27*

LNUs (*n* = 78)	High-dependency demand
London – Whittington	224.3
Nottingham – City	205.0
London – Isleworth	201.5
London – Leystonstone	201.1
London – King's	182.0
London – Lewisham	171.6
London – Croydon	169.7
Oxford	159.9
Bristol – Royal	157.6
Romford	149.5
Hull	149.3
Oldham	146.9
Leicester – Royal	146.8
Norwich	145.5
Northampton	139.8
Burnley	137.0
Dewsbury	136.0

continued

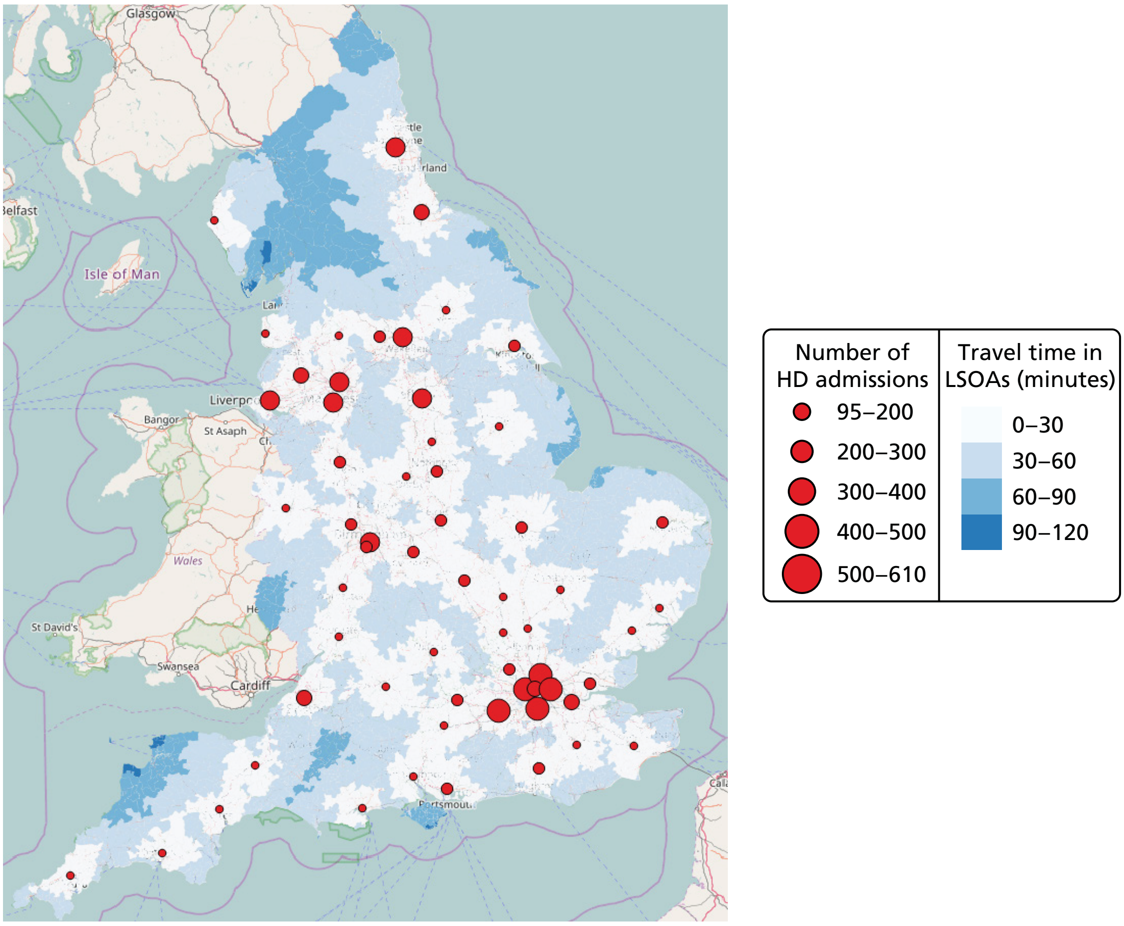
TABLE 23b Configuration of LNUs relating to *Figure 27* (*continued*)

LNUs (n = 78)	High-dependency demand
Manchester – North	135.4
London – St Peter	134.1
Wirral	134.0
Poole	126.6
Swindon	126.5
Redhill	125.1
Walsall	124.5
Uxbridge	124.4
Coventry	123.3
Birmingham – Edgbaston	121.6
Sheffield	116.2
Birmingham – Children's	111.1
London – St George	109.7
Ashford	108.6
Colchester	108.0
Worcester	105.7
Truro	102.4
Stevenage	102.2
Ashington	101.6
Ipswich	94.3
Sunderland	93.5
Wigan	93.1
Barnsley	91.7
Shrewsbury	90.9
Harlow	89.2
York	88.3
Tunbridge Wells	87.2
Ashton under Lyne	87.2
Sutton Coldfield	86.7
Taunton	85.7
Lincoln	83.7
Hastings	83.4
Exeter	81.2
Durham	81.2

TABLE 23b Configuration of LNUs relating to *Figure 27* (*continued*)

LNUs (*n* = 78)	High-dependency demand
Birmingham – City	81.1
Winchester	79.6
Doncaster	79.6
Kettering	78.9
Basingstoke	78.7
Lancaster	76.2
Darlington	75.2
Crewe	73.7
Whitehaven	73.7
Blackpool	72.5
Nuneaton	72.5
Margate	70.1
King’s Lynn	68.3
Dorchester	67.2
Ormskirk	60.0
Great Yarmouth	59.5
Torbay	58.8
Worksop	47.5
Macclesfield	46.4
Hereford	45.1
Scunthorpe	44.5
Stafford	43.8
Chichester	43.7
Harrogate	43.6
Barnstaple	39.4
Keighley	39.3
Newport, Isle of Wight	30.7

(a)



(b)

Average travel time	21 minutes
Maximum travel time	99 minutes
Mothers within 30 minutes of closest NICU	82%
Minimum number of HD admissions	96
Maximum number of HD admissions	610

FIGURE 28 Example of optimal configuration with 30 NICUs and 30 LNUs with high-dependency demand (*Tables 24a* and *b*). HD, high dependency. (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); and (b) key performance indicators.

TABLE 24a Configuration of NICUs relating to *Figure 28*

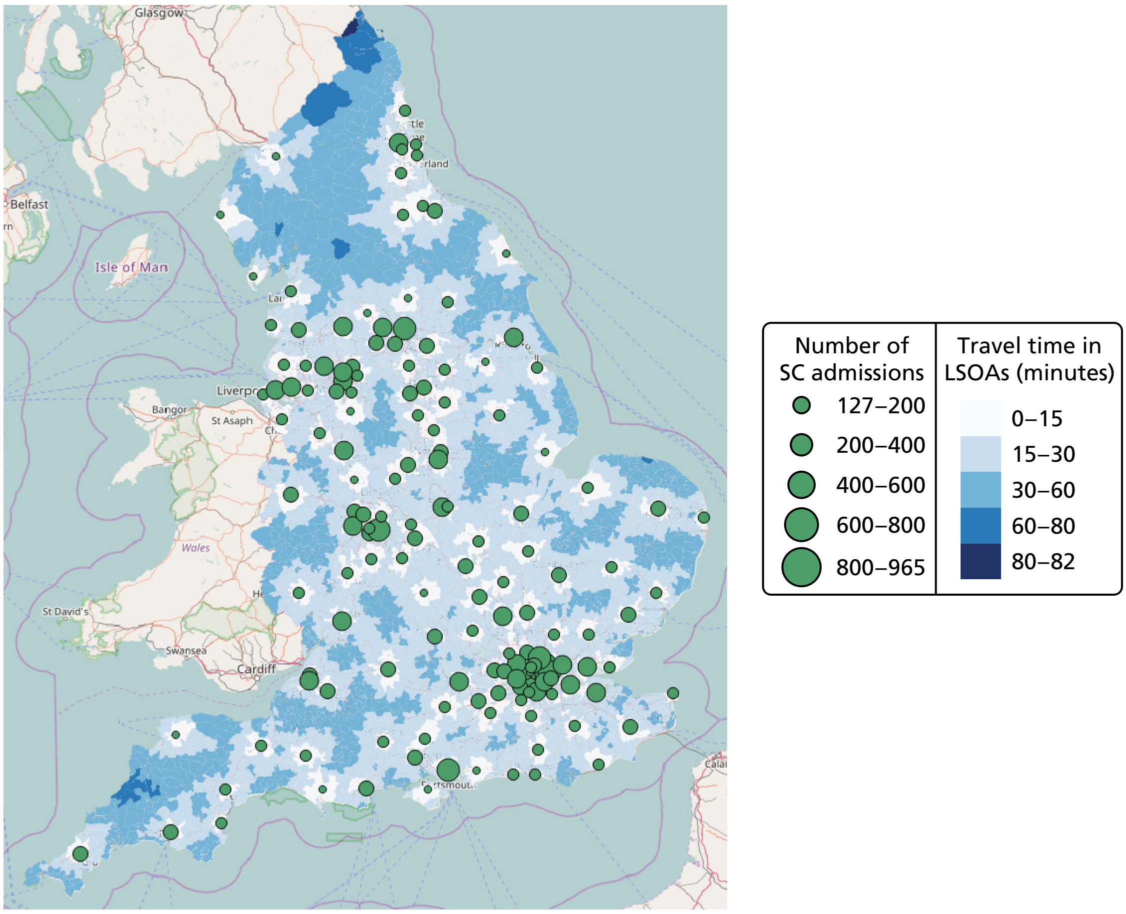
NICUs (n = 30)	High-dependency demand
London – Edmonton	568.6
London – Queen C	522.7
London – St Peter	522.6
London – Croydon	520.1
Leeds	476.1
Manchester – North	470.3
Birmingham – Children's	452.1
Rotherham	446.9
Newcastle upon Tyne	435.1
Liverpool	417.4
Dartford	370.5
Bristol – Royal	359.2
London – University College London	306.3
Stockton-on-Tees	305.4
Basildon	299.1
Wolverhampton	298.2
Haywards Heath	296.5
Watford	274.2
Portsmouth	268.5
Coventry	249.6
Hull	241.3
Nottingham – University	239.2
Stroke-on-Trent	223.8
Norwich	222.5
Southampton	182.2
Burnley	171.3
Swindon	156.4
Bedford	153.6
Taunton	121.9
Plymouth	116.7

TABLE 24b Configuration of LNUs relating to *Figure 28*

LNUs (n = 30)	High-dependency demand
London – Newham	609.8
Manchester – University	406.4
Wigan	304.7
Bradford	251.8
Birmingham – Edgbaston	218.9
Peterborough	213.0
Reading	209.6
Northampton	209.0
Leicester – Royal	207.7
Ashford	194.8
Luton	186.7
Oxford	184.5
Derby	180.7
Basingstoke	177.2
Exeter	170.2
Poole	164.8
Cambridge	163.4
Gloucester	157.8
Blackpool	157.2
Sutton-in-Ashfield	152.8
Worcester	142.8
Colchester	141.2
Stevenage	138.6
Tunbridge Wells	138.1
Lincoln	113.7
York	112.3
Truro	105.0
Shrewsbury	101.9
Whitehaven	97.2
Ipswich	95.7

Access to special care

(a)



(b)

Average travel time	14 minutes
Maximum travel time	82 minutes
Mothers within 30 minutes of closest NICU	95%
Minimum number of SC admissions	127
Maximum number of SC admissions	965

FIGURE 29 Current configuration with 45 NICUs, 78 LNUs and 38 SCUs with special care demand (*Tables 25a–c*). SC, special care. (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); and (b) key performance indicators.

TABLE 25a Configuration of NICUs relating to *Figure 29*

Existing NICUs (n = 45)	Special care demand
Leeds	964.6
Birmingham – Heartlands	831.0
Portsmouth	813.1
Bradford	769.0
Bolton	711.2
Newcastle upon Tyne	702.9
Bristol – Royal	702.5
Gillingham	661.5
Luton	660.6
Manchester – St Mary	659.3
Leicester – Royal	640.7
London – King’s	634.2
Stroke-on-Trent	633.2
Nottingham – University	613.2
Liverpool	611.3
Burnley	608.5
Hull	600.3
Norwich	586.6
Oldham	578.5
Coventry	549.7
Oxford	528.4
Southampton	523.0
London – Queen C	520.9
London – Royal	512.0
London – Homerton	510.7
Nottingham – City	503.7
Preston	502.0
Plymouth	497.8
Birmingham – Children’s	489.5
Ashford	484.3
Bristol – North	482.9
London – St George’s	480.8
Middlesbrough	479.7
Sheffield	479.3
London – St Peter’s	476.3

TABLE 25a Configuration of NICUs relating to *Figure 29* (*continued*)

Existing NICUs (*n* = 45)	Special care demand
Wolverhampton	464.9
Birmingham – Edgbaston	453.9
Cambridge	428.2
Wirral	395.5
Brighton	372.7
Sunderland	346.9
Stockton-on-Tees	333.6
London – Guy's and St Thomas'	291.4
London – Chelsea	279.1
London – University College London	185.0

TABLE 25b Configuration of LNUs relating to *Figure 29*

Existing LNUs (*n* = 78)	Special care demand
London – Edmonton	958.3
London – Newham	829.6
London – Leystonstone	794.1
London – Northwick Park	785.4
Prescot	731.2
London – Lewisham	676.5
London – Croydon	676.2
Romford	666.3
Basildon	648.3
Dudley	639.1
Reading	625.1
Manchester – North	603.5
Gloucester	601.9
Slough	580.7
London – Barnet	562.7
Swindon	556.8
Walsall	554.9
Peterborough	547.6
Rotherham	540.7
Poole	532.6
Derby	527.0
Uxbridge	513.8

continued

TABLE 25b Configuration of LNUs relating to *Figure 29* (*continued*)

Existing LNUs (*n* = 78)	Special care demand
Bath	511.5
Northampton	509.7
Milton Keynes	492.9
Kingston upon Thames	487.6
Manchester – University	475.5
Colchester	463.2
Halifax	459.3
Truro	456.4
London – Queen E	455.9
Stevenage	452.8
Pontefract	446.8
London – Whittington	445.2
Camberley	411.3
Shrewsbury	405.3
Barnsley	400.0
Harlow	397.6
London – St Helier	393.4
Watford	392.3
Sutton-in-Ashfield	391.9
Redhill	381.6
Ashton under Lyne	375.2
Wigan	375.0
Warrington	373.1
Exeter	362.1
Birmingham – City	361.7
Doncaster	354.9
Ipswich	346.8
Tunbridge Wells	346.5
Chelmsford	346.4
Kettering	345.4
Basingstoke	344.4
Stockport	337.3
Aylesbury	336.1
Taunton	326.6
Blackpool	323.1

TABLE 25b Configuration of LNUs relating to *Figure 29* (*continued*)

Existing LNUs (*n* = 78)	Special care demand
Worcester	321.0
Lincoln	307.2
Southend	303.5
Crewe	300.5
King's Lynn	296.8
York	283.5
Ormskirk	267.6
Grimsby	261.8
Chesterfield	261.7
Salisbury	230.7
Winchester	230.2
Chester	226.5
Lancaster	221.7
Burton upon Trent	208.4
Scunthorpe	198.5
Chichester	189.5
Macclesfield	178.9
Keighley	175.0
London – St Mary	152.1
Dorchester	150.6
Newport, Isle of Wight	137.1

TABLE 25c Configuration of SCUs relating to *Figure 29*

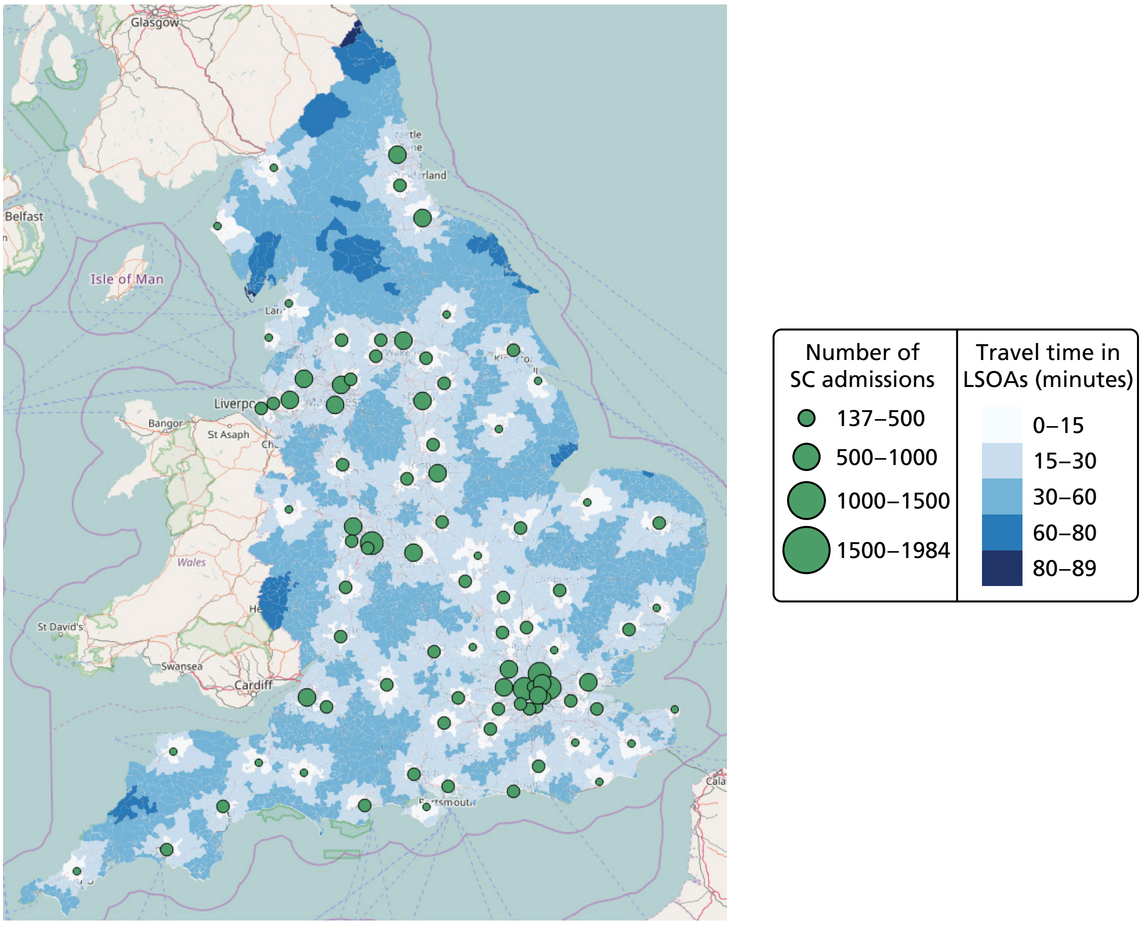
Existing SCUs (*n* = 36)	Special care demand
London – Isleworth	772.0
Dartford	673.7
Dewsbury	421.2
London – Royal Free	378.1
Sutton Coldfield	378.0
Hastings	367.5
Durham	355.3
Guildford	342.6
Redditch	326.8
Darlington	324.7
Nuneaton	323.0
Margate	312.5

continued

TABLE 25c Configuration of SCUs relating to *Figure 29* (*continued*)

Existing SCUs ($n = 36$)	Special care demand
Worthing	307.3
Haywards Heath	303.3
Orpington	295.5
Bedford	291.1
Bury Saint Edmunds	283.8
Warwick	269.9
Leicester – General	268.6
Great Yarmouth	265.4
Ashington	262.6
Torbay	262.0
Yeovil	245.0
Gateshead	235.3
London – Epsom	230.0
Huntingdon	224.4
South Shields	220.1
Worksop	209.4
Hereford	201.2
Boston	197.2
Carlisle	194.9
Scarborough	193.5
Stafford	193.5
Harrogate	184.5
Banbury	184.0
Barnstaple	175.7
Whitehaven	146.9
Barrow-in-Furness	127.0

(a)



(b)

Average travel time	17 minutes
Maximum travel time	89 minutes
Mothers within 30 minutes of closest NICU	89%
Minimum number of SC admissions	137
Maximum number of SC admissions	1984

FIGURE 30 Example of optimal configuration with 30 NICUs, 30 LNUs and 30 SCUs with special care demand (*Tables 26a–c*). SC, special care. (a) Background map © OpenStreetMap contributors;[60] the data are available under the Open Database Licence and the cartography is licenced as CC BY-SA (www.openstreetmap.org/copyright); and (b) key performance indicators.

TABLE 26a Configuration of NICUs relating to *Figure 30*

NICUs (n = 30)	Special care demand
Birmingham – Children's	1984.1
London – Edmonton	1852.4
London – Queen C	1537.7
Leeds	1435.0
Newcastle upon Tyne	1419.5
Rotherham	1388.2
Basildon	1323.8
Manchester – North	1295.9
Bristol – Royal	1175.4
Stockton-on-Tees	1114.8
Coventry	1107.3
Nottingham – University	1066.4
Watford	1034.5
Wolverhampton	1007.1
Stroke-on-Trent	993.4
Portsmouth	953.9
Dartford	936.0
Norwich	907.7
London – Croydon	875.4
Southampton	756.5
Burnley	746.2
London – St Peter	744.2
Hull	723.4
London – University College London	686.8
Bedford	662.5
Liverpool	611.3
Haywards Heath	594.3
Swindon	580.4
Plymouth	517.5
Taunton	337.6

TABLE 26b Configuration of LNUs relating to *Figure 30*

LNUs (n = 30)	Special care demand
London – Newham	1629.1
Manchester – University	1448.1
Wigan	1063.5
Bradford	929.2
Leicester – Royal	902.2
Derby	805.3
Reading	734.7
Peterborough	724.5
Luton	715.2
Birmingham – Edgbaston	710.2
Gloucester	703.5
Sutton-in-Ashfield	681.3
Oxford	678.1
Cambridge	642.4
Northampton	621.0
Colchester	618.6
Exeter	603.9
Worcester	575.8
Poole	572.4
Stevenage	552.9
Basingstoke	546.3
Ashford	484.3
Blackpool	465.2
Truro	456.4
York	454.4
Shrewsbury	445.7
Ipswich	426.8
Lincoln	394.6
Tunbridge Wells	362.8
Whitehaven	160.1

TABLE 26c Configuration of SCUs relating to *Figure 30*

SCUs (n = 30)	Special care demand
Uxbridge	1491.2
Prescot	1166.5
London – King's	1090.2
London – Homerton	1001.7
London – Lewisham	981.6
Oldham	947.0
Kingston upon Thames	819.3
London – St Helier	748.0
Durham	743.6
Dudley	684.0
Gillingham	661.5
Worthing	639.5
Wirral	614.1
Guildford	592.8
Pontefract	581.0
Halifax	576.5
Bath	525.4
Doncaster	522.4
Harlow	439.7
Aylesbury	405.1
Yeovil	388.3
Hastings	372.0
Kettering	359.3
Lancaster	341.5
Grimsby	321.8
Margate	312.5
King's Lynn	304.6
Carlisle	199.6
Barnstaple	175.7
Newport, Isle of Wight	137.1

Appendix 2 Mortality model

In this appendix, we report the analysis of causal effect of high-volume units for infants at a gestational age of between 26 and 32 weeks. We present the results of estimating a model of exposure to NICUs at birth or within 48 hours of birth. We present the results of a third, additional, analysis comparing clinical outcomes between NICUs, LNUs and SCUs using travel time to the closest LNU and SCU.

Analysis of causal effect of high-volume units for infants born between 26 and 32 weeks of gestational age

Sensitivity analyses were conducted on mortality estimates for infants born between 26 and 32 weeks of gestational age. Following the advice of policy-makers, infants born at < 26 weeks were excluded from the estimates in the sensitivity analysis because it was felt that they included a subgroup of infants whose chances of survival were low, making them more likely to die at the hospital of birth.

The model estimated the effect of high-volume units that treat ≥ 100 infants born weighing < 1500 g per year. The model used an IV approach using travel time as the instrument and covariates were included for gestational age, gestational age squared, birthweight, birthweight squared, the sex of the infant, the lowest decile of the IMD score of the mother's residence, the mode of delivery and the number of foetuses.

Table 27 summarises the results of the sensitivity analysis and shows that excluding infants born at < 26 weeks of gestational age halves the mortality effect of birth in high-volume units compared with other units (linear SMM 0.02 vs. 0.05 percentage points in all the infants of < 32 weeks of gestational age; IV bivariate probit 0.6 vs. 1.2 percentage points in all the infants of < 32 weeks of gestational age). The instrument remains strong in this analysis and, as before, the Hausman test statistic confirms that treatment variable (delivery in a high-volume hospital) is endogenous (tested using the Hausman test statistic having a *p*-value of < 0.01). As anticipated, the reduced number of deaths in this sensitivity analysis leads to an inevitable loss of power, particularly in the linear SMM model (IV linear SMM, $p = 0.269$; IV bivariate probit, $p = 0.108$).

TABLE 27 Causal effect on mortality of birth in high-volume units for infants born at 26–31 weeks of gestational age ($n = 10,821$)

Outcomes and test statistics	LPM	IVs linear SMM	IV bivariate probit model (marginal effect)
Birth at high volume, absolute risk difference (SE)	0.016** (0.005)	–0.020 (0.018)	–0.006 (0.004)
Instrument strength for minimum travel time (minutes) to high-volume hospital, coefficient (SE)	N/A	–0.003*** (0.000)	–0.018*** (0.001)
F-/*z*-test statistic	N/A	28.8	28.7
Hausman test χ^2 statistic of H0: no endogeneity of treatment variable	N/A	N/A[a]	9.1***

*$p < 0.10$, **$p < 0.05$, ***$p < 0.01$.
H0, null hypothesis; N/A, not applicable; SE, standard error.
a This model was estimated using the Generalised Method of Moments and so the Hausman test is not applicable.
Note
Controlled covariates: age and age squared at birth, birthweight and birthweight squared, sex, deprivation of residence, mode of delivery and fetus number.

Additional analysis of clinical outcomes of exposure to a neonatal intensive care unit at birth or within 48 hours of birth

In our main IV analysis, we found that NICU level had no effect on neonatal mortality relative to mortality in non-NICU hospitals of birth. We were therefore interested to know whether or not the lack of effect had anything to do with the compensating effects of transfers, which in our data set took place for 1694 infants. Of these, 1519 had data on transfer time available, and 990 (65%) had a recorded first transfer to a NICU from a lower level hospital of birth taking place within 48 hours of delivery. Thus, we investigated the effect of exposure to a NICU within 48 hours of birth in our original sample of infants born at < 32 weeks of gestation and in the subset of those born at < 32 weeks after excluding those born at < 26 weeks. The results are presented in *Tables 29* and *30*, and suggest that exposure to a NICU because of transfers had no role in the lack of effect of exposure to a NICU on mortality. These results should be interpreted with caution because our instrument may have limited ability to control for confounding associated with neonatal transfers within 48 hours of delivery.

Additional analysis comparing clinical outcomes between neonatal intensive care units, local neonatal units and special care units using travel time to the closest local neonatal unit and special care unit

Birth in a hospital with a NICU does not appear to result in any difference in terms of mortality relative to other hospitals. These results are based only on using one instrument: travel time to closest hospital. In this appendix, we present additional analyses of the effect of hospital designation on mortality using additional instruments in the form of travel time to the closest LNU and SCU.

A simultaneous equations multivariate probit model was adopted to implement an IV model of neonatal mortality as a function of unit level of birth (considering SCU, LNU and ICU). This model uses the three available instruments represented by the travel time to the closest hospital for each of the three unit levels.

Table 30 summarises the results of the IV model on the marginal effect (first column) and the results using a naive probit model of the same mortality function (second column). In each of these two models, covariates were included for gestational age, birthweight, sex of infant, delivery mode, number of foetuses and the lowest decile of the IMD score of the residence of the mother.

TABLE 28 Causal effect on mortality of birth in NICU for infants born at 26–31 weeks of gestational age ($n = 12,687$)

	Causal effect on mortality of neonatal care in hospitals with a NICU		
Outcomes and test statistics	**LPM**	**IVs linear SMM**	**IV bivariate probit model (marginal effect)**
Birth at NICU, absolute risk difference (SE)	–0.007 (0.005)	–0.014 (0.016)	–0.003 (0.011)
Instrument strength for minimum travel time (minutes) to tertiary-level hospital, coefficient (SE)	N/A	–0.008*** (0.000)	–0.022*** (0.001)
t-/*z*-test statistic	N/A	30.4	33.0
Hausman test χ^2 statistic of H0: no endogeneity of treatment variable	N/A	N/A[a]	0.3

**p* < 0.10, ***p* < 0.05, ****p* < 0.01.
H0, null hypothesis; N/A, not applicable; SE, standard error.
a This model was estimated using the Generalised Method of Moments and so the Hausman test is not applicable.
Note
Controlled covariates: age and age squared at birth, birthweight and birthweight squared, sex, deprivation of residence, mode of delivery and foetus number.

TABLE 29 Causal effect on mortality of exposure to neonatal care at tertiary units within 48 hours of birth: excluding births at < 26 weeks of gestational age ($n = 10{,}821$)

	Causal effect on mortality of neonatal care in hospitals with a NICU		
	LPM	IVs linear SMM	IV bivariate probit model (marginal effect)
Birth at NICU, absolute risk difference (SE)	0.007* (0.004)	–0.006 (0.013)	–0.003 (0.008)
Instrument strength for minimum travel time (minutes) to tertiary-level hospital, coefficient (SE)	N/A	–0.008*** (0.000)	–0.023*** (0.001)
t-/*z*-test statistic	N/A	29.3	32.6
Hausman test χ^2 statistic of H0: no endogeneity of treatment variable	N/A	N/A[a]	1.54

*$p < 0.10$, **$p < 0.05$, ***$p < 0.01$.
H0, null hypothesis; N/A, not applicable; SE, standard error.
a This model was estimated using the Generalised Method of Moments and so the Hausman test is not applicable.
Note
Controlled covariates: age and age squared at birth, birthweight, birthweight squared, sex, deprivation of residence, mode of delivery and fetus number.

TABLE 30 Causal effect on mortality of birth in lower-level hospitals (LNUs and SCUs) relative to NICU ($n = 11{,}037$)

Output metric	Naive univariate probit regression	IV multivariate probit model (marginal effect)
Birth in a LNU, absolute risk difference (SE)	0.004 (0.005)	0.019** (0.009)
Birth in a SCU, absolute risk difference (SE)	0.016* (0.009)	0.004 (0.013)
Instrument strength		
Minimum travel time (minutes) to NICU, coefficient (SE)	N/A	LNU equation: 0.032 (0.001)***
		SCU equation: 0.015 (0.001)***
Minimum travel time (minutes) to LNU, coefficient (SE)	N/A	LNU equation: –0.053 (0.001)***
		SCU equation: 0.006 (0.001)***
Minimum travel time (minutes) to SCU, coefficient (SE)	N/A	LNU equation: 0.007 (0.001)***
		SCU equation: –0.063 (0.002)***
Likelihood-ratio test statistic	N/A	46.9***
Hausman test *z*-statistic of H0: no endogeneity LNU treatment variable	N/A	2.3**
Hausman test *z*-statistic of H0: no endogeneity SCU treatment variable	N/A	1.5
Test *z*-statistic H0: valid over-identifying restriction of minimum travel time to NICU	N/A	0.5

*$p < 0.10$, **$p < 0.05$, ***$p < 0.01$.
H0, null hypothesis; N/A, not applicable.
Note
Controlled covariates: age and age squared at birth, birthweight, birthweight squared, sex, deprivation of residence, mode of delivery and fetus number.

The naive probit model suggests that birth in a SCU is associated with an increased risk of neonatal death relative to birth in a NICU (1.6 percentage points of absolute risk difference), whereas no difference is apparent between birth in a LNU compared with birth in a ICU. However, the opposite result is found with the IV model, in which birth in a LNU exposes infants to additional neonatal death risks (absolute risk difference 1.9 percentage points), whereas no difference is observed between birth in a SCU and a NICU. It must be noted that, in accordance with the IV model diagnostic statistics, the hypothesis that birth in a SCU is exogenous cannot be rejected, which suggests that a simpler model with IVs applied only to the LNU treatment variable may be enough to validly estimate causal effects. At the bottom of *Table 30*, the non-significant *z*-test statistic of 0.5 does not lead to rejection of the hypothesis that the extra (over-identifying) instrument of minimum travel time to a NICU estimates the same causal effect as the other two instruments. These additional analyses that were undertaken suggest that the NICU does appear to reduce mortality compared with the other levels of care, by 1–2%, and so suggests that NICUs in themselves have some beneficial impact on mortality compared with other levels of care.

Appendix 3 Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram (outlining literature search)

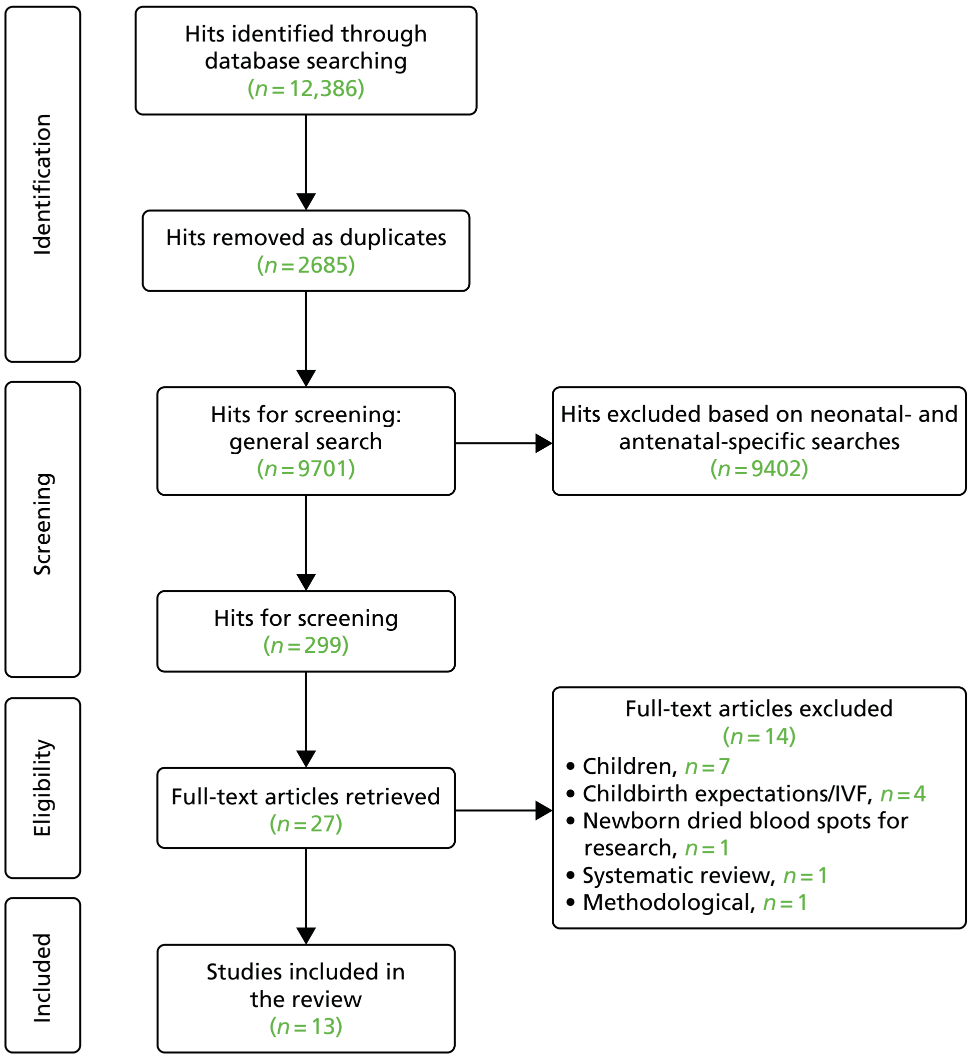


FIGURE 31 The PRISMA flow diagram. Reproduced with permission from Moher *et al.*[109] This is an Open Access article distributed in accordance with the terms of the Creative Commons Attribution (CC BY 4.0) license, which permits others to distribute, remix, adapt and build upon this work, for commercial use, provided the original work is properly cited. IVF denotes in vitro fertilisation.

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