Identifying exceptional cystic fibrosis care services: combining statistical process control with focus groups

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Scientific summary

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Background

Cystic fibrosis (CF) is a common genetic disease affecting approximately 9000 people in the UK. People with CF are treated at specialist centres that offer dedicated paediatric or adult care.

The CF Trust – the national UK charity for CF – maintains a national registry database (UK CF Registry) of detailed clinical data on all patients attending these specialist centres. Patients are invited to attend annual reviews at their specialist centre and, based on these data, the Trust produces annual reports describing the health of patients.

Since 2008, the annual reports published by the CF Trust have included comparisons between centres in terms of key clinical outcomes, using simple rankings. Although these comparisons give a sense of the distribution of outcomes between centres, they encourage the reader to assume, for example, that centres with the highest values of lung function measure are 'better' than those with lower values. This is misleading because the rankings make no allowances for differences in patient case mix and there are no formal tests comparing centres.

An alternative – and increasingly common – approach is to use statistical process control (SPC) charts. These charts were developed as a means of studying the variability of 'processes' over time and have been shown to be useful when comparing health-care providers, as well as the outcomes of single providers, over time. For the charts, summary outcome measures for individual units are plotted against time or the size of the unit, with control limits [at either 2 or 3 standard deviations (SDs)]. They are designed to highlight any variability caused by factors outside the process, referred to as 'special-cause variation'. Centres whose summary measures are outside the control limits (either higher or lower) are said to exhibit special-cause variation.

Aims and objectives

The aim of this project was to determine whether or not there are statistically meaningful differences in key health outcomes between CF centres in the UK and to determine the processes of care driving such differences.

Our objectives were to:

1. establish a framework to allow future comparisons of key measures of quality in terms of care structures and processes
2. develop statistical models using data from the national CF Registry that allow us to adjust clinical outcomes for the patient case mix at centres
3. compare centres using SPC charts to determine whether or not important differences exist between centres on key clinical outcomes
4. consult with experts in CF care to turn existing care guidelines into maps of the structures, processes and policies (SPPs) required to deliver good outcomes and to identify factors that could facilitate or hinder these from being successful
5. consult with CF patients to ascertain what patients believe are the factors that could facilitate or hinder these SPPs from being successful
6. collect data from individual centres and describe the structures and processes of care at these centres based on the SPP mapping exercise
7. explore how the SPPs of care differ between centres with exceptional outcomes and the remaining centres.

Methods

There were a number of components to this work, each of which is described below.

Centre comparisons

We used annual review data (2007–15, inclusive) from the CF Registry to study the following outcomes.

For paediatric centres:

- forced expiratory volume in 1 second (FEV_1) at age 12 years (2007–15)
- FEV_1 at age 15 years (2007–15)
- FEV_1 change from ages 13 to 15 years (2007–12)
- body mass index (BMI) percentile at age 15 years (2007–12).

For adult centres:

- FEV_1 change from ages 18 to 21 years (2007–12)
- BMI change from 18 to 21 years as described above (2007–12).

To inform the analyses of these age groups and to confirm any trends observed, we also conducted single-year analyses (2007–15, inclusive) of FEV_1 using CF Registry data of patients of all ages.

For each outcome, we first generated a funnel plot (form of SPC chart) for the outcomes unadjusted for case mix. We used 99.8% (3 SDs) and 95% (2 SDs) control limits to identify special-cause variation but focused our attention on centres that were outside the 3-SD limits in all of our analyses. We then adjusted our outcomes for different patient case-mix characteristics. Multilevel models allowing for clustering by centre were used for this adjustment and, from these case-mix adjusted models, new funnel plots were generated.

From these analyses, we also explored trends in intravenous (i.v.) antibiotic usage in terms of the proportion of patients on treatment and the median number of days on treatment.

Consultations to identify factors that would help to understand variability

Clinicians

Focus groups were held with clinicians from adult and paediatric centres so that the key determinants of quality of care could be understood. The purpose of the workshops was to explore the factors that contributed to high-quality care with the intention of developing a questionnaire that could be sent to CF centres to reveal local patterns of care. The questionnaires were disseminated online and centre directors of all of the specialist paediatric and adult centres were invited to participate. They were also asked to share the questionnaire with members of their multidisciplinary team (MDT) so that we might receive responses from a range of clinicians.

Patients

We conducted a focus group with the CF Trust's team of clinical care patient advisors. The first part focused on the participants' own experiences of CF care. In the second part, participants were asked to consider, from their own experiences, factors that could facilitate or hinder the success of the SPPs that were identified by clinicians and for their views on the importance of these clinician-defined SPPs.

Results

Centre comparisons

The analyses showed that using funnel plots on an important and well-completed clinical outcome such as FEV_1 allows us to identify some trends in special-cause variation. We described our results using anonymised identification (ID) numbers for the centres; these ID numbers do not correspond to the codes currently used by the CF Trust in its reports.

Centre comparisons: paediatrics

We observed that centre 39 was frequently an outlier, with FEV_1 levels outside the lower 3-SD limit in our unadjusted analyses of young adults. It also repeatedly showed special-cause variation in our year-by-year analyses, along with centres 12 and 52 (outside the lower 3-SD limits) and centres 25, 28 and 31 (outside the upper 3-SD limits).

The first step in the pyramid of investigation model involves checking the data. We noted that data were relatively complete for the case-mix variables considered, but some centres (39 and 43) had lower than average rates of complete FEV_1 data among those patients having annual review encounters.

Following the pyramid of investigation approach, we found that a check of the case mix revealed some differences in the patient populations at these sites. Adjustment for age, for example, brought centre 28 within the 3-SD control limits, despite patients at this centre being only slightly younger than the mean. Centres 12 and 31 were brought within the control limits after adjustment for other case-mix factors. We observed that centre 12 had more female patients and more patients who had a G551D mutation, as well as having fewer patients with the most deprived socioeconomic status. Centre 31 had fewer patients who were homozygous for the DF508 mutation and more who were pancreatic sufficient than the overall paediatric patient population.

After case-mix adjustment, three of the original six centres showed patterns of special-cause variation in FEV_1: centres 39 and 52 (lower than average FEV_1 values) and centre 25 (higher than average FEV_1 values). We next considered care; an objective and routinely collected measure of CF care is the use of i.v. antibiotics, which has been shown to be related to better clinical outcomes. We noted that there was a tendency for centre 25 – which had better FEV_1 outcomes – to be near or outside the lower control limits for the proportion of patients on i.v. antibiotics. Conversely, centres 39 and 52, which had poorer FEV_1 outcomes, tended to have higher than average proportions of patients on i.v. antibiotics. This, however, tells us about receiving treatment, not about needing treatment.

Centre comparisons: adults

As performed in the paediatric analysis, we studied FEV_1 in many different manners from decline in early adulthood to studying all adults in repeated single-year analyses. In analyses unadjusted for case mix, we observed that centres 9, 7 and 34 were frequently outside the 3-SD limits and centre 40 appeared outside the limits in 2007.

Following the pyramid of investigation, we found that adjustment for case mix is essential to avoid misattributing differences in outcomes to care or missing important differences. Adjustment for age, for example, brought centre 40 from within the 3-SD control limits to outside these limits from 2009 to 2015 (except in 2012).

We then considered other patient case-mix characteristics. In models adjusted for a wider range of case-mix variables, the adjustment substantially affected centre 40 only, which was brought within the 3-SD limits. This centre differed from the average in many ways: the centre had fewer patients who were homozygous for DF508, more with the milder G551D mutation and more who were pancreatic sufficient. Our findings at the other centres (9, 7 and 34) could not be attributed to case mix. To gauge whether or not the patterns observed were related to the use of multilevel modelling for our case-mix adjustment, we tried a fixed-effects

modelling approach, which suggested that case mix alone might not explain the outlying results at centre 40.

We then compared the centres in terms of the proportion of patients receiving i.v. antibiotics. Surprisingly, centres 9 and 40 frequently exhibited special-cause variation in this variable but in different 'directions', and not entirely consistently with our hypothesis. For example, both centres had fewer patients on i.v. antibiotics but one had better FEV_1 outcomes than the average and the other had poorer FEV_1 outcomes than the average. Exploring the number of days on i.v. antibiotics proved challenging as patterns were not consistent and the high level of skewness of this variable means that standard funnel plots – and the associated control limits – would not provide reliable estimates.

Consultation with clinicians

Focus groups with clinicians from adult and paediatric centres were conducted to identify what they felt were the important SPPs for delivering good CF care. There was a focus on adequate staffing and sufficient time with different members of the MDT; monitoring and measuring of clinical outcomes and treatment adherence; communication within the MDT and with patients; patient education and support; and standard policies on how to deal with infection control and eradication, as well as changes in lung function and BMI. From this work, questionnaires for the directors of adult and paediatric CF centres were developed and disseminated nationally. It was hoped that the results from these questionnaires could help to inform conversations with CF centres that had outcomes that were better or worse than the average. The response rate, however, was relatively poor; this made it difficult to draw inferences or to use the responses to start a dialogue with centres.

Consultation with patients

A focus group with the CF Trust's team of expert patient advisors was conducted to identify what patients thought was necessary for good CF care. Based on this, we devised a questionnaire to be disseminated to patients nationally. The focus group concentrated on the characteristics of three areas: (1) good inpatient stays, (2) good outpatient visits and (3) well-functioning CF units. In relation to inpatient stays, the group highlighted a number of issues, including the importance of patient involvement in devising a clear action plan and self-management of medication during the stay. Access to an appropriate and sufficient number of members of the MDT and consistent quality of care over the course of a 1-week stay were also identified as important. In the outpatient setting, patients felt that it was important to have sufficient time to see all members of the MDT, to have access to an outpatient pharmacy and to speak to an experienced consultant who has detailed knowledge of the patient. It was felt that a well-functioning CF unit will educate patients about CF, develop a constructive dialogue between patients and CF unit staff, have patients involved in the measurement and management of key aspects of CF and provide consistent access to the unit's MDT. Owing to time constraints, we were unable to conduct the patient survey within the time frame of this study, but it will take place subsequently.

Conclusions

These findings confirm that the annual review data can be used to identify differences in clinical outcomes between centres and that case-mix characteristics might explain some of these differences. However, this adjustment does not account for all differences; therefore, further work is needed to explore the results that were obtained.

Strengths and limitations

This study's greatest strength is the completeness and coverage of the data source. Close to 100% of CF patients consent to their data being included in the CF Registry and, in later years of analysis, the proportion of registered patients having an annual review recorded in a given year approached 90%. We also consulted with the CF clinical community and patients in order to understand what outcomes should be studied for our work to be relevant.

We tried different strategies in our data analysis – from considering different ways of adjusting for age to using different reference equations for FEV_1 – and we have largely drawn the same conclusions. However, despite the large number of data collected, we were not always able to adjust for all relevant case-mix characteristics as a result of data not being available (such as characteristics at transition to adult care among older adults). In addition, although the CF Registry is very complete, some centres are much smaller than others and we cannot exclude the possibility that we have failed to detect special-cause variation in these very small centres.

The low response rates to our centre director surveys meant that we were unable to get a clear sense of the structures and processes of care at all centres. We did get supplemental information from the CF Registry – such as days on i.v. antibiotics – but this does not cover the full picture of how care is structured and delivered at a centre.

Future work

This work has raised questions that could be addressed in future work, for example 'How does the antibiotic treatment affect outcomes?'. This would be helpful if explored in discussions with sites at which we could discuss the approaches to treatment of CF patients in different scenarios.

Another key question is 'How do patients feel about the care that they receive?'. We developed a detailed questionnaire for patients that we did not have the time to disseminate and analyse within the context of this grant.

Implications for practice

The learning from this work regarding the importance of case-mix adjustment, the limitations of such plots of detecting special-cause variation in small centres, the usefulness of making comparisons across years rather than using single-year analyses and the potential usefulness of incorporating available process measures can all be incorporated into future practice at the CF Registry.

Our approach provides a framework for future centre comparisons in CF or other disease areas for which routine clinical data are available on well-defined patient populations. These can usefully form the basis of discussions with sites about outcomes – subject to the limitations described here – when following the pyramid of investigation approach.

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