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Additional therapy for young children with spastic cerebral palsy: a randomised controlled trial

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Abstract

Additional therapy for young children with spastic cerebral palsy: a randomised controlled trial

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Objectives: To investigate whether, in the short and medium term, additional support by (a) a physiotherapy assistant improved physical function in young children with spastic cerebral palsy and (b) a family support worker improved family functioning.
Design: This was a multi-centre randomised controlled trial (RCT) with blinded assessments and a cost-effectiveness analysis. The children studied had spastic cerebral palsy that was the consequence of perinatal adversity. All were less than 4 years old on entry to the study.
Setting: In the child development centre and in the home.
Participants: Seventy-six families completed the intervention period. Forty-three families were reassessed 6 months after the end of the intervention and 34 of these after a further 6-month period.
Interventions: Randomisation was to: (a) a group who received extra physiotherapy from a physiotherapy assistant; (b) a group who received standard physiotherapy; and (c) a group where the child received standard physiotherapy and the family was also visited by a family support worker. Children in all groups continued to receive standard physiotherapy in addition to the study interventions.
Main outcome measures: The child outcome measures were motor functioning, developmental status and adaptive functioning. The family outcome measures were self-reported maternal stress, level of family needs and parental satisfaction.
Results: There was no evidence that additional physical therapy for 1 hour per week for 6 months by a physiotherapy assistant improved any child outcome measure in the short or medium term. Intervention by a family support worker did not have a clinically significant effect on parental stress or family needs. Over the 6-month period the total cost of services for each child ranged from £250 to £6750, with higher costs associated with children with more severe impairments. No significant relationship was found between measures of intensity of services received by the children and families and the main outcome measures. Low-functioning children, in terms of both motor and cognitive function, were more likely to receive more services in terms of range and frequency. Parents generally reported high satisfaction ratings after all interventions and some stated that the interventions had benefited the child and/or the family. There was therefore a discrepancy between the perceptions of these parents and the objective, quantitative measurements. The family support workers identified a small number of families who were experiencing considerable family problems, but who had not been referred for appropriate support by any other agency.
Conclusions: The findings of this study provide support for the current literature that there was no evidence that additional intervention (in this case by a physiotherapy assistant or family support worker) helped the motor or general development of young children with spastic cerebral palsy. Nor was there any quantitative evidence that providing extra family support helped levels of parental stress and family needs. The implication was that the provision of extra physical therapy does not necessarily improve the motor function of a young child with cerebral palsy and additional family support should not automatically be assumed to be beneficial. In addition, no significant association was found between the intensity of the local services provided and any outcome measure, other than a slight association with lowered family needs. The provision of local services was related to the severity of the child's impairments and not to family difficulties. A small group of families with complex family problems needed more service input. There was a wide range in

the costs of services. Research is needed to examine what 'sufficient' levels of provision or therapy might be for which children and which families. A time series of different levels of input and outcomes would provide valuable information for practitioners. It is also recommended that future assessments of therapies of this type adopt a similar multifaceted approach, which is likely to be more suitable than a simple RCT for the evaluation of clinical interventions where the effects are complex. The most appropriate measures of outcome should be used, including assessment of provision of information and emotional support for families.

Contents

List of abbreviations

ANCOVA	analysis of covariance
BLCS	Brief Locus of Control Scale
CDC	child development centre
CG	control group
CI	confidence interval
COPE	an inventory to assess coping strategies
CP	cerebral palsy
CQ	cognitive quotient
DL	Daily Living (scale)
DQ	developmental quotient
FACES	Family Adaptability and Cohesion Evaluation Scale
FNS	Family Needs Scale
FSS	Family Support Scale
FSW	family support worker
FSWG	family support worker group
GHQ	General Health Questionnaire
GMFM	Gross Motor Function Measure
HOME	Home Observation for Measurement of the Environment
ICER	incremental cost-effectiveness ratio
LOC	Locus of Control
MQ	motor quotient
NDT	neurodevelopmental therapy
PA	physiotherapy assistant
PAG	physiotherapy assistant group
PSI	Parental Stress Index
RCT	randomised controlled trial
SD	standard deviation
SfP	School for Parents
T1	pre-intervention assessment
T2	post-intervention assessment
T3	first follow-up, 6 months after T2
T4	second follow-up, 12 months after T2

All abbreviations that have been used in this report are listed here unless the abbreviation is well known (e.g. NHS), or it has been used only once, or it is a non-standard abbreviation used only in figures/tables/appendices in which case the abbreviation is defined in the figure legend or at the end of the table.

Executive summary

Objectives

It has been suggested that children with cerebral palsy should not only have their physical needs addressed, but also that there should be support for the family.

This study separated these functions by investigating whether in the short and medium term additional support by (a) a physiotherapy assistant improved physical function in young children with spastic cerebral palsy and (b) a family support worker improved family functioning; children in all groups received standard physiotherapy in addition to the study interventions. In addition, the study examined the needs of the families and the factors affecting child and family functioning in relation to services received and outcome.

Design

This was a multi-centre randomised controlled trial (RCT) with blinded assessments and a cost-effectiveness analysis. The children studied had spastic cerebral palsy that was the consequence of perinatal adversity. All were less than 4 years old on entry to the study.

Randomisation was to: (a) a group who received extra physiotherapy from a physiotherapy assistant; (b) a group who received standard physiotherapy; and (c) a group where the child received standard physiotherapy and the family was also visited by a family support worker. Children in all groups continued to receive standard physiotherapy in addition to the study interventions.

Both quantitative and qualitative methods were used in this trial.

Participants

Seventy-six families completed the intervention period. Forty-three families were reassessed 6 months after the end of the intervention and 34 of these after a further 6-month period.

Main outcome measures

The child outcome measures were:

- motor functioning (Gross Motor Function Measure)
- developmental status (Griffiths Mental Developmental Scales)
- adaptive functioning (Vineland Scales).

The family outcome measures were:

- self-reported maternal stress (Parent Stress Index)
- level of family needs
- parental satisfaction.

Results

The RCT found that:

- There was no evidence that additional physical therapy for 1 hour per week for 6 months by a physiotherapy assistant improved any child outcome measure in the short or medium term.
- Intervention by a family support worker did not have a clinically significant effect on parental stress or family needs.
- Over the 6-month period the total cost of services for each child ranged from £250 to £6750, with higher costs associated with children with more severe impairments.

The multivariate analyses found that:

- There was no significant relationship between measures of intensity of services received by the children and families and the main outcome measures.
- Low-functioning children, in terms of both motor and cognitive function, were more likely to receive more services in terms of variety and frequency.

The qualitative analysis found that:

- Parents generally reported high satisfaction ratings after all interventions and some stated that the interventions had benefited the child

and/or the family. There was therefore a discrepancy between the perceptions of these parents and the objective, quantitative measurements.
- The family support workers identified a small number of families who were experiencing considerable family problems, but who had not been referred for appropriate support by any other agency.

Conclusions

The findings of this study provide support for the current literature that there was no evidence that additional intervention (in this case by a physiotherapy assistant or family support worker) helped the motor or general development of young children with spastic cerebral palsy. Nor was there any quantitative evidence that providing extra family support helped levels of parental stress and family needs. The implication was that the provision of extra physical therapy does not necessarily improve the motor function of a young child with cerebral palsy and additional family support should not automatically be assumed to be beneficial. In addition, no significant association was found between the intensity of the local services provided and any outcome measure, other than a slight association with lowered family needs.

The provision of local services was related to the severity of the child's impairments and not to family difficulties. A small group of families with complex family problems needed more service input.

There was a wide range in the costs of services.

Implications for health care

Physical therapy services were largely child focused. No evidence of significant provision of family support was found, other than in one area that offered wider support in facilitating contact between services and referral to other services. The qualitative methodology showed that there were some families which benefited from the family support worker intervention. These families of children with cerebral palsy had little input from other sources. They were experiencing high stress levels and had high levels of unmet needs. Support for these families was at a relatively superficial level and there was no indication of support by social or psychological support, that is, the focus appeared to be on physiotherapy services directed towards the child.

It appears that more funds are not needed. However, families who might particularly benefit from family-focused intervention were not appropriately targeted.

There was some evidence of over-provision and/or of a sub-optimal service mix for some of these children and their families. There should be specific focus on avoiding duplication and defining the criteria that are used to decide on service provision for individual children and their families.

If the physiotherapist is to be the key professional (key worker) for a child with cerebral palsy, guidance should be given on how to explain to parents that more intensive or more frequent physical therapy may not necessarily be warranted. A key worker for the child with cerebral palsy and his or her family needs to understand what family support entails, and their role in providing it and acting as gatekeepers for referral to other agencies. If this really is an important part of the paediatric physiotherapist's role, appropriate training and resources need to be provided.

Recommendations for future research

Research is needed to examine what 'sufficient' levels of provision or therapy might be for which children and which families. Key issues are:

- How the allocation of resources to individual children and families is decided
- The variability among child development centres in relation to how families are assessed, the formulation of a family plan, referrals to other agencies and interagency working. One approach might be to compare the effectiveness of a service with a key worker who has clear management protocols and develops an individualised care plan with the present less structured approach.

A time series of different levels of input and outcomes would provide valuable information for practitioners.

Various methodologies were used for this study. It is recommended that future assessments of therapies of this type adopt a similar multifaceted approach, which is likely to be more suitable than a simple RCT for the evaluation of clinical interventions where the effects are complex. The most appropriate measures of outcome should be used, including assessment of provision of information and emotional support for families.

Chapter 1
Introduction

Cerebral palsy (CP) affects about two children in every 1000 and is one of the most common acquired causes of disability of central nervous system origin. Children with CP are routinely referred for physical therapy. However, there is confusion and debate in the research and clinical literature about the efficacy of treatments and about the relationship between the intensity of treatment and consequent benefits. In the field of early intervention for children with disabilities, the last two decades have seen an evidence-supported shift from condition-focused to child-focused to family-focused interventions. An investigation of the effectiveness of the services for both child and family functioning with children with CP was therefore warranted.

Children with CP comprise a heterogeneous group. Although all are affected by a non-progressive movement disorder, the pattern of an individual's disability varies with maturation of the central nervous system. Disability can range from very severe tetraplegia, where all four limbs are seriously affected by spasticity, to relatively mild diplegia, where the legs are stiffer than the arms. There may be hypotonicity or ataxia; there may or may not be seizures and impaired vision and hearing; associated cognitive impairment is variable, but can be profound. There is an association between CP and prematurity and therefore likely to be a preponderance of socially deprived families. Twins and higher multiples are more likely to be affected than singletons.[1] Therefore, since children with CP and their families constitute a heterogeneous population, any relevant research should consider such diversity.

The main focus of management for children with CP is their physical disability. A key contact point with the health services for the family is therefore the physiotherapist, a professional person who has been trained to concentrate on the child's physical needs and to enable that child to make the most of his or her physical resources. In the UK, physiotherapists who treat children with CP generally use an eclectic form of therapy known as neurodevelopmental therapy (NDT). It was broadly based on the work of Bobath.[2] The philosophy was that movements can be learned, that normal posture was needed for normal movements and that parents should be involved to ensure that therapy was applied consistently and frequently.

There was uncertainty about how often physical therapy should be carried out and a perception among some parents and professionals that increasing the frequency of physiotherapy was likely to be beneficial. This results in a demand for higher levels of provision, with consequent cost implications for the service and for the family in terms of time, emotion and finance. At present, the supervising physiotherapist decides on the frequency with which therapy is applied and whether and how to support the family by taking into account the perceived needs of the child and local resources, but there was little guiding evidence.

Although physiotherapists may concentrate on movement facilitation, they frequently find that they become general advisers to the family. This wider role is not one for which they have been specifically trained, nor is it one for which they are likely to have sufficient time. However, if it is accepted that the development of the child will be affected by environmental factors, this supportive role, which may be beneficial, needs to be investigated. The study was designed to evaluate these two functions separately. The study included an investigation of the services that each child received and a cost evaluation of the interventions and of the services generally. The study also aimed to investigate the relationships between the child with CP, the family, community services and the factors that influence these interactions.

Physical therapy for children at risk of developing CP or with established CP

In spite of the relatively high incidence of CP and the widespread use of physiotherapy for children with this condition, there have been comparatively few intervention studies.

Physical therapy for children at risk of developing CP

Turnbull[3] published a meta-analysis of studies undertaken between 1973 and 1993 for babies

born at risk of developing CP. She concluded that there was no unequivocal evidence that intervention before the development of abnormal physical signs produced an advantage in motor development in children who later developed CP. Her conclusions were supported by three subsequent studies using NDT for children considered to be at high risk in South Africa,[4] Montreal[5] and Liverpool.[6] All failed to show an effect. The one study that looked at the effect of physiotherapy intervention for children at risk for the development of CP and the prevalence of depression in their mothers failed to find any positive benefit.[7] These studies also demonstrated the difficulty in predicting CP reliably.

Physical therapy for children with established CP

Various treatments have been used to manage children with CP, including surgery, orthoses, drugs and physical therapy/physiotherapy. Physical therapy is the most common intervention. There are various types of physical therapy, such as sensory integrative therapy, progressive pattern movement, the Vojta method, neuromuscular facilitation, conductive education, biofeedback and behaviour modification techniques,[8] but most physiotherapists in the UK practise NDT. Studies fall into three categories: those that compared NDT with other therapeutic approaches; those that evaluated the effectiveness of NDT, often comparing them with broader stimulation methods; and those that looked at the intensity and nature of NDT.

NDT compared with other physical therapies

The advantages of one particular therapeutic method over another have been difficult to establish through scientific enquiry. In an early review, Harris[9] concluded that there were too few well-controlled studies to provide firm conclusions. Hur[8] reviewed 37 studies that considered the outcome of physical therapeutic interventions for children with CP between 1966 and 1994. Only seven studies used randomisation or a control or comparison group.[10–16] The studies were small, with samples ranging from 12 to 47 children. The period of intervention varied between 4 weeks[14] and 4 years.[15] There was also wide variation in frequency (intensity) of physiotherapy provided. The four larger studies[10,13–15] together included 151 children, and failed to detect any differences between groups. Hur concluded that because of the methodological shortcomings of small sample sizes (limiting the ability to detect a treatment effect), short-term interventions, poorly controlled experimental conditions and lack of follow-up, no conclusions could be reached on the efficacy of any of the physical therapies.

Effectiveness of NDT

Bower and McLellan[17] evaluated eight major studies of physiotherapy undertaken between 1962 and 1993 on children with an established diagnosis of CP.[10–12,16,18–21] They too considered that methodological shortcomings prevented any definite conclusions being drawn about the effectiveness of physiotherapy. In addition to Hur's criticisms, they observed that there was a lack of objective, valid and reliable outcome measures and a lack of power calculations. Furthermore, they argued that grouping subjects together raised a possibility that important changes for individuals would be missed.[17]

One notable study of the effect of NDT on motor outcome was by Palmer and colleagues in 1988.[20,22] This high-quality randomised trial compared physical therapy alone with physical therapy combined with a programme of infant stimulation, comprising motor, sensory, language and cognitive activities. A strength of this study was that the pattern of disability of the subjects was homogeneous: all children studied were between 12 and 19 months old and all had spastic diplegia. Forty-eight infants were randomly assigned to receive either 12 months of physical therapy or 6 months of a programme of infant stimulation followed by 6 months of physical therapy. There were no significant differences between the groups in the incidence of contractures or the need for orthopaedic interventions. After the intervention, infants who had received physical therapy alone progressed more slowly than those who received the programme of infant stimulation followed by physical therapy. On the Griffiths Development Test, they had lower mean mental quotient (66 versus 76, $p = 0.05$) and locomotor quotient (49 versus 58, $p = 0.02$) and were less likely to walk. These differences persisted after 12 months of therapy. The addition of infant stimulation for 6 months had a better effect than NDT alone, underlining the importance of monitoring other inputs from local services and of measuring outcomes that are broader than locomotion alone.

Intensity of NDT

In spite of the lack of evidence for a consistent effect of NDT, many parents and professionals believe that the therapy is effective for children with CP and, in many cases, that the more intensively it is applied (i.e. the more sessions), the

better.[23] Again, available studies are equivocal and have methodological shortcomings. In 1997, using a randomised cross-over design, Law and colleagues[24] found that intensive NDT (45 minutes of therapy twice per week and 30 minutes per day at home) had no benefit when compared with regular NDT (therapy between once per week and once per month) and a 15-minute programme at home three times per week.

In 1998, Reddihough and colleagues[25] found that young children (12–36 months old, mean age 22 months) with CP involved in conductive education, one of the most intensive physical therapies available, made similar progress to those given traditional NDT.

In contrast, in a series of studies, Bower and colleagues[23,26] used intensive physiotherapy and intervention periods ranging from 2 to 5 weeks and detected improvement in motor function on the Gross Motor Function Measure (GMFM). The largest study, with 44 children, used a 2-week intervention period and contrasted goal setting and intensive physiotherapy in a 2 × 2 design. They found a clinically and statistically significant effect for the group with set goals and a non-significant trend for intensive physiotherapy.[26] None of these studies followed the children post-intervention to determine long-term effects. Bower and colleagues repeated the study in 2001 (after the present study commenced) with 56 children in a randomised design.[23] In their study, no significant effects were found. There was a trend for benefit for the intensive group of 2.5% points on the GMFM only when additional covariates were introduced, not in the primary analysis. Any benefit had disappeared at the 6-month follow-up.

In conclusion, many of the available studies had methodological problems, particularly with sample size and power, use of controls, heterogeneity of samples, range of potentially confounding variables and looking at persisting effects of the intervention. Hence it was not possible to draw confident conclusions about the efficacy of physical therapy for children with cerebral palsy.

Family-focused interventions

Mothers of preterm infants, many of whom may be considered to be 'at risk' for CP, are much more likely to come from social classes 3–5 than from 1 or 2,[6] and there is a pervasive influence of psychosocial adversity on the mother's mental health.[7] Recognition of this has prompted a shift from medical models to social models of disability, that is, from organic to child-focused approaches and from focus on the treatment of children and their disability to a broader focus on the family and immediate environment – the ecological model. This shift has been supported by several studies of early intervention.[27–30] For example, Seitz and colleagues[30] provided a programme to enhance the abilities of mothers of families with social disadvantage to help themselves and their children. This produced mothers who were more able and whose children at 10 years of age outperformed control children on a variety of school and adaptive measures.

Parental participation in early intervention programmes for children with learning difficulties and social disadvantage is well established.[31,32] It has been argued that such involvement increases parents' understanding of their child's development and capacities, and helps them to develop appropriate expectations regarding their child's future.[20]

The importance of considering the social context when studying the effectiveness of therapies with children with physical disability was illustrated by a study by Sloper and Turner in 1992.[33] They investigated the service needs of 107 families of children under 8 years of age who were unlikely to walk without aids; 40% had CP and 34% were under 3 years of age. Although there was an average of 68 service contacts per year by 10 different professionals, there was still an association between high stress levels in mothers and high rates of unmet needs. The mothers lacked information about their child's condition, what services were available and how to access them. They needed help with their child's mobility and housing adaptation. Families likely to require substantial service support were those where the father was unemployed, where there were more stressful life events, where the child had associated cognitive impairment and where the mother used a passive coping strategy. As found by studies of people with other disabilities,[34] the authors concluded the main need was for a key worker with a broader role than is normally delivered by a specific therapist.

The diversity of services provided and the interrelationship between many influential variables were demonstrated in a large multivariate American investigation of child development and family adaptation.[27] A total of 199, 1-year-old children with disabilities (40% with

motor impairment) were enrolled in an early intervention programme. Support services were heterogeneous in terms of both contacts with different professional agencies and whether they were home or centre based. A major finding was that the more severe the child's disability was, the more intense (frequent and longer) were the service contacts. The duration of the visits and whether they were at home were associated with decreased parental stress over the 12-month study period. Nevertheless, severity of motor impairment was the most important predictor of child development and was associated with negative family outcomes. There was lower maternal adjustment to the child's disability in families of children with relatively severe impairment.

Hence some children are 'doubly vulnerable' and at risk from both biological and environmental factors.[27,35] Stressors created both by family burdens and by disability interact within the family to produce an additional effect on child development. Consequently, it has been suggested that family needs should be assessed individually and that intervention programmes should be needs based and tailored specifically towards family characteristics and stressors.[17,33,36]

Early intervention programmes are considered capable of altering these non-optimal family interaction patterns directly or by moderating the impact of stressors that influenced those patterns.[28] In 2001, Ketelaar and colleagues[37] reviewed studies that examined parental involvement in programmes for children with CP. They found that few studies had been explicitly designed to investigate parental involvement and that programmes differed in content, objectives, nature and degree and duration of parental participation. However, most reported positive results and recommended that parents should be included in goal setting and that programmes should be adapted to families' capabilities and resources.

In 1991, Davis and Rushton developed a model programme for family-centred early intervention in the UK using a parent adviser.[38] This involved a comprehensive assessment of the family based on a resource-needs model. Although this was shown to be effective with socially disadvantaged ethnic minority families with young children with disability,[39] the benefits were less clear for those who were not immigrants or from ethnic minorities. Like many studies, it did not take into account the levels of service being provided to families.

However, the Avon study in 1998[40] targeted 'at-risk' preterm infants below 33 weeks' gestation to evaluate the effectiveness of a home-based structured developmental programme (Portage) compared with family support using the parent adviser model. No significant effects were found. The Portage programme appeared to result in a small advantage [+4.3 general quotient points, 95% confidence interval (CI) 1.6 to 7.0] compared with social support by a parent adviser (+3.4 general quotient points, 95% CI 1.4 to 6.1), particularly for the smallest infants (birth weights of less than 1250 g) and those with brain injuries. Social variables confounded the results and locomotor subscales were not reported. Nevertheless, general family support seemed to be as effective as the child-focused intervention.

Studies of the cost-effectiveness of physiotherapy with young children with CP

A literature search of the NHS Centre for Reviews and Dissemination databases [Database of Abstracts of Reviews and Effects (DARE), NHS Economic Evaluation Database (NHS EED) and the Health Technology Assessment (HTA) Database] was conducted at the start of the study (1997). Entering the keywords 'cerebral palsy' and 'cost'; 'cerebral palsy' and 'economic' revealed no papers on the economics of physiotherapy for young children with CP, highlighting the need for the current economic evaluation.

Since starting the project, two relevant studies have been done. They focused on young adults, rather than young children, and both took into account a range of disabilities which included cerebral palsy. Neither assessed physiotherapy. In 2002, Bent and colleagues[41] applied a cost–consequence analysis from an NHS perspective as part of this trial on a sample of young people aged between 17 and 28 years in four centres around the UK. The trial compared a team-based approach versus *ad hoc* health services in a retrospective cohort study. The results revealed that the young adult team-based approach cost an average of £650 per person and the *ad hoc* service group cost an average of £798 per person.

Beecham and colleagues conducted a study in 2001 estimating the cost consequences of supporting young adults aged between 18 and 25 years with hemiplegic CP.[42] They calculated the total cost and the additional costs accrued to the

public and independent sector due to their participants' disabilities. They found that the group of 81 subjects studied cost a little over £1 million to support during a 1-year period, 43% of which was related to their impairments. People with a combination of associated conditions, such as intellectual impairment, were found to have costs nearly 50 times greater than those of a patient with simple hemiplegia.

Conclusions and rationale for the present study

Although the belief persists that more intense physiotherapy is important for children with CP, the evidence is equivocal. This impacts on the role of the physiotherapist and whether they try to meet a demand for more intense physiotherapy for children or adopt a broader, more family-focused, role. Previous studies can be criticised on a number of methodological grounds, including small and heterogeneous samples, short-term interventions, failure to randomise groups, lack of follow-up, a restricted range of measures and failure to research the impact on families. There was also a lack of information about the range, distribution and costs of services. Hence a larger controlled study was necessary.

Aims of the study

1. To evaluate the effect of increasing the frequency of NDT.
   One approach would have been for children to be given extra treatment by their treating physiotherapist. However, this was rejected, for the following reasons. First, it would have been costly to increase the input of physiotherapists who were responsible for treatment. Second, these practitioners were mostly based in child development centres (CDCs) and there are simply not enough physiotherapists to undertake such additional work. Third, involving a different physiotherapist at the child's home in addition to the treating physiotherapist would have been likely to cause confusion and add a confounding variable to the study. Fourth, it was argued that someone other than a highly qualified and relatively expensive physiotherapist could provide much of the additional hands-on therapy and encourage parents to use the same techniques. The approach adopted was therefore to increase the intensity of physiotherapy by appointing a physiotherapy assistant (PA), who visited the child at home and acted under the instruction of the supervising physiotherapist who devised the goals for the programme.
2. To examine the belief that the effect of physical therapy on the child is not just through physical support but also through supporting the family.
   The two functions of a physiotherapist – physical therapy and family support – were separated. The PA provided an input that was targeted at improving physical outcome, and a family support worker (FSW), based on the parent adviser model[38,39] and supervised by a clinical psychologist), would meet any family needs.
3. To assess the cost-effectiveness of the intervention against a broader evaluation of service costs.
4. To provide information about the way in which families with children with CP functioned, the amount of services received and the factors that determined this.

The following hypotheses were addressed:

1. Extra home-based physical therapy by a PA improves motor function in the child with spastic CP.
2. Extra home-based physical therapy by a PA improves general development of the child with spastic CP.
3. Extra home-based intervention by an FSW improves the functioning of the family of a child with spastic CP.

Chapter 2
Methods

Design

This main part of the study was designed as a randomised controlled trial (RCT) with blinded assessment. Both quantitative and qualitative measures were used. Eligible infants were randomly allocated to one of three groups: a physiotherapy assistant group (PAG), a family support worker group (FSWG) or a control group (CG).

Health economic costing information was collected about the services received, their impact on young children with CP and their families.

In addition, quantitative and qualitative methodologies were used to collect information about the children, their families and the services used. This was to (a) examine how the intervention fitted into the context of the families' lives, (b) enhance knowledge about the nature of development of young children with CP and (c) gain information about the needs and adaptation of their families, with a view to informing social policy.

Ethical approval

The study was started before the Multicentre Research Ethics Committee system was established and ethical approval was obtained from all the localities where the study was undertaken, that is, from the relevant Local Research Ethics Committees.

Inclusion and exclusion criteria

Children were eligible if they had CP of perinatal origin that was predominantly spastic in type. A standard definition of CP was used: a persistent disorder of movement and posture caused by non-progressive defects or lesions of the immature brain.[43] Children were excluded from this study if the brain damage that caused CP occurred after they were 6 months old or if the predominant pattern of CP was of dystonia or ataxia. Children with CP were entered if they were less than 3 years old at the start of this study. Later, to improve recruitment, the age for entry was increased to less than 4 years.

Recruitment

Once the diagnosis had been disclosed to parents, children were referred to the study either by their paediatrician or by the senior paediatric physiotherapist in the CDC. A few parents self-referred in response to posters in toy libraries, health centres or CDCs.

Most parents were asked if they would be willing to consider taking part in this study by their supervising physiotherapist. If they were, the research coordinator was informed and she arranged to meet the parents and explain the study and gave parents an information sheet. Parents then had 1 week to consider whether they wished their child to be entered into the study.

It was initially planned to undertake the study in a geographically defined area, comprising Merseyside, Cheshire and North Wales. In this area there are 35,000 births annually and, using an incidence of CP of two per 1000 births, around 65 new cases of CP would be expected each year. Taking into account the exclusion criteria and delays in diagnosis for less severely affected children, we anticipated around 40–45 potential referrals each year. Unfortunately, while the present study was being set up, two large centres responsible for the care of disabled children in Liverpool and South Sefton became committed to a study of botulinum toxin and were therefore unable to participate. This reduced the potential referrals by around 40%. Recruitment was monitored as the study progressed. Several changes were made to improve recruitment. These included sending reminders to physiotherapists and paediatricians, who were also invited to annual seminars on progress, increasing the referral age to less than 4 years and seeking referrals from a wider geographic area, including Greater Manchester and Lancashire.

The centres involved were Liverpool, Wirral (Arrowe Park Hospital), Chester, Warrington, St Helen's (Whiston Hospital), Ormskirk, Wigan

and Leigh, Crewe, Wrexham, Widnes, Manchester and Rhyl.

The interventions

Families in the CG received physiotherapy and support in the routine manner used in their CDC. This was a study of current practice. No attempt was made to standardise the style of physiotherapy given. Nevertheless, all the physiotherapists in the CDCs involved in this study carried out therapy in a similar fashion, namely NDT, as described in Chapter 1.

In addition to their regular physiotherapy, the families in the PAG received one extra session of physiotherapy lasting for around 1 hour each week from a PA. For most, this was in their homes, but occasionally at the CDC that the child attended regularly. Initially, the PA attended the CDC with the child or attended a joint home visit with the paediatric physiotherapist responsible for the child's therapy and who supervised the PA throughout the intervention. The PA was instructed by the physiotherapist about the approach and aims for that child. Thus, the intervention was focused on the needs and goals for the individual child. When the PA visited the child's home, she also encouraged the parents to continue with any advice given by the CDC physiotherapist in treating the child. The intervention period was for 6 months.

Families allocated to the FSWG had weekly home visits by an FSW, which, like those in the PAG, lasted around 1 hour over a 6-month intervention period. The FSWs were parents of children with CP and had attended a short course for parent advisers.[39] This consisted of 12 weekly sessions, each 3 hours long, run by two clinical psychologists. Talks and practical exercises were used that focused on family and child needs and interpersonal and counselling skills. The FSW began by discussing family needs and how she could best help the family. The ethos was of an equal partnership with joint decision-making about family needs and how they might be met. The FSWs had fortnightly meetings with a clinical psychologist for supervision.

Randomisation

Children were randomised using a minimisation technique[44] by a telephone call to an independent statistician on a remote site. Randomisation was stratified to take account of some of the variations between children with CP and their families: (1) maternal education, (2) pattern of spasticity and (3) the geographical area where the child's treatment was based.

1. Maternal education has been shown to be a determinant of child development; it is often associated with other family and nurturance variables and was likely to be a factor in the ability to mobilise community services.
2. The pattern of CP is likely to be associated with developmental progress and response to intervention. A standard definition of spasticity was used:[1] tetraplegia denoted the involvement of all limbs with the arms being equally or more affected than the legs; diplegia was used when the legs were more severely affected than the arms; hemiplegia was used when one side of the body only was involved, with the arm more affected than the leg.
3. Support services were likely to vary between geographical areas and so area (i.e. CDC) was viewed as an important ecological variable.

Assessment measures

The measures used are listed in *Table 1*. They comprised a broad range of methodologies: standardised scales, self-completed questionnaires, observations and interviews.

Appendix 1 describes these measures and their selection, validation and reliability in detail.

Descriptive independent variables

Demographic and other descriptive data were collected from information supplied by the referring paediatrician or physiotherapists and from the initial interviews with the families conducted by the project coordinator, a senior physiotherapist, who also observed the child. The coordinator confirmed the coding of the pattern of disability.

Two quotients were calculated for the purposes of this study as indicators of the child's cognition: cognitive quotient, (CQ) and motor development, motor quotient (MQ). These were based on the relevant subscales of the Griffiths Mental Development Scale. Details are provided in Appendix 1.

Amongst the variables likely to influence the effects of any intervention are the nature and intensity of additional therapeutic, educational or

TABLE 1 *Measures used*

Descriptive variables	Mediating variables	Outcome variables
Child characteristics	**Intervention**	**Child competence**
Age, gender, birth order	Intervention group	Motor functioning (GMFM)
Gestation		Development (Griffiths)
Birth weight	**Other services**	Adaptive functioning (Vineland)
Ethnicity	Family support (FSS)	
Age of entry into study	Additional therapy	**Parent/family**
Age of diagnosis		Parenting Stress (PSI) or GHQ
Cognitive status (CQ)	**Family ecology**	Family needs
General health	Quality of home (HOME)	Parental satisfaction
Natural/fostered/adopted	Family cohesion (FACES)	
Severity of motor impairment (MQ)	Family adaptability (FACES)	
Pattern of CP	Coping (COPE)	
	Locus of Control (LOC)	
Family characteristics		
Maternal and paternal ages		
Relationship status		
Maternal education (years of)		
Socio-economic status		
Maternal and paternal		
educational qualifications		
occupation/employment status		
general health		
mental health		
Siblings/family size		
Area of residence		

formal support received by families. Several sources were used to collect information about these:

- The initial interview.
- Daily diaries kept by the parents of contacts with service personnel.
- Structured interviews about contacts with services that were completed by the independent assessor at the beginning and end of the intervention period.
- Diaries kept by the PAs and FSWs.
- A standardised questionnaire, the Family Support Scale (FSS), which assessed families' perceptions of formal and informal support. It was sent to families before the first assessment, completed by the parents and collected at the assessment.
- A structured questionnaire sent by post at the end of the study to ask about services received and their frequency.

Family ecology

Four standardised scales were used to measure the quality of the child's family environment:

- An independent assessor (see below) completed the Home Observation for Measurement of the Environment (HOME)[45] at each assessment.
- Parents completed the Family Adaptability and Cohesion Evaluation Scale (FACES III) (Olson DH: personal communication, 1995).[46] This resulted in two factors, Family Cohesion and Family Adaptability.
- Parents completed the Coping with Stress Scale,[47] which provided three factors, Active Coping, Adaptive Coping and Maladaptive Coping, the last of which essentially focused on passive mechanisms.
- Parents also completed the short-form Locus of Control (LOC) Scale.[48]

Outcome variables – child measures

The assessments were done in the family home by an independent senior paediatric physiotherapist, who was blind to the group allocation. She carried out assessments on four occasions:

- T1 – pre-intervention assessment, prior to the intervention period
- T2 – post-intervention assessment, 6 months later (after the intervention/observation period)
- T3 – first follow-up, 6 months after T2
- T4 – second follow-up, 12 months after T2.

Gross motor ability

The main outcome measure to assess the effects of additional physiotherapy was the GMFM.[49–54]

Developmental status
The full raw score of the Griffiths Mental Development Scale was used.[55]

Adaptive functioning
The raw scores of the Daily Living (DL) and Socialisation domains of the Vineland Adaptive Behaviour Scales[56] were used to assess any generalised effects of the intervention.

Outcome measures – family and parent
The parents completed three questionnaires:

1. The Parenting Stress Instrument (PSI).[57–58] This provided two major measures, Parent Domain and Child Domain, and also a score indexing potential stress from Life Event Stressors. The Parent Domain scores measured stress from factors related to parenting and the Child Domain measured factors directly related to the child. The former was used as the main outcome measure.
2. The General Health Questionnaire (GHQ).[59] The GHQ measured mental health (anxiety, depression and social isolation) and provided an indicator of general stress.
3. Family Needs Scale (FNS).[60]

Both the PSI and GHQ are well standardised measures. The FNS has not been standardised but was developed for use in early intervention programmes.

Parent satisfaction with the intervention was assessed by an interview at the end of the intervention period (see Appendix 1 for protocol and rating).

Economic measures
These are described in detail in the section 'Cost-effectiveness analysis' (p. 26).

Sample size – power analysis
The primary outcome for this study was improved motor function for these children with spastic CP. The degree of improvement considered clinically important was an improvement in the grade of severity. Data from the GMFM showed that for children aged less than 3 years with CP, an improvement from a 'moderate' to 'mild' group was demonstrated by an increase of 14 points on the total GMFM score.[49] To achieve 80% power with 5% significance required 51 children in each group.

Based on this analysis, we intended to recruit 180 children, i.e. 60 in each group. This would have allowed for a mean difference of at least seven points to be detected on the Vineland Adaptive Behaviour Scale. We had expected these numbers to be achievable because previous experience had led us to anticipate that at the start of the project there would be at least 50 existing children with CP who would be able to be randomised immediately, in addition to those identified as having developed CP during the course of the study.

Procedure
A description of the steps in the recruitment process and the investigation is set out in *Figure 1*. The main procedures were as follows:

1. Letters with information sheets were sent to paediatricians and physiotherapists at participating CDCs.
2. The physiotherapist referred families who were willing to be contacted and provided details of the child's condition, gender, address and telephone number.
3. The family was contacted by the project coordinator, who carried out a home visit and gave a fuller explanation of the study. Child and family demographic details were collected. Consent forms were signed and the 'cooling off' period and withdrawal explained.
4. These details were sent for randomisation.
5. At the end of the cooling off period, the coordinator informed the independent assessor and sent the appropriate questionnaires to the family for completion and collection by the assessor.
6. The independent assessor arranged a home visit. She completed the assessment of the child and family, explained the diaries and collected the questionnaires. She was not aware of the group to which the family had been allocated when she did the baseline assessment. At later assessments, parents were reminded not to discuss group allocation with the assessor.
7. Families were informed as to which group they had been allocated. (a) If in the Family Support Group, the FSW made contact by telephone and arranged the first visit. (b) If in the Physiotherapy Assistant group, the PA contacted the child's physiotherapist and arranged to meet for discussion of programme and arranged the first home visit.
8. Near the end of the intervention, the PA or FSW informed the project coordinator, who arranged for the independent assessor to visit. The project coordinator kept records of starting and completion dates and the number of home visits made.
9. Post-intervention assessment followed within 4 weeks of the end of the intervention or, in the case of the CG, the 6-month period.

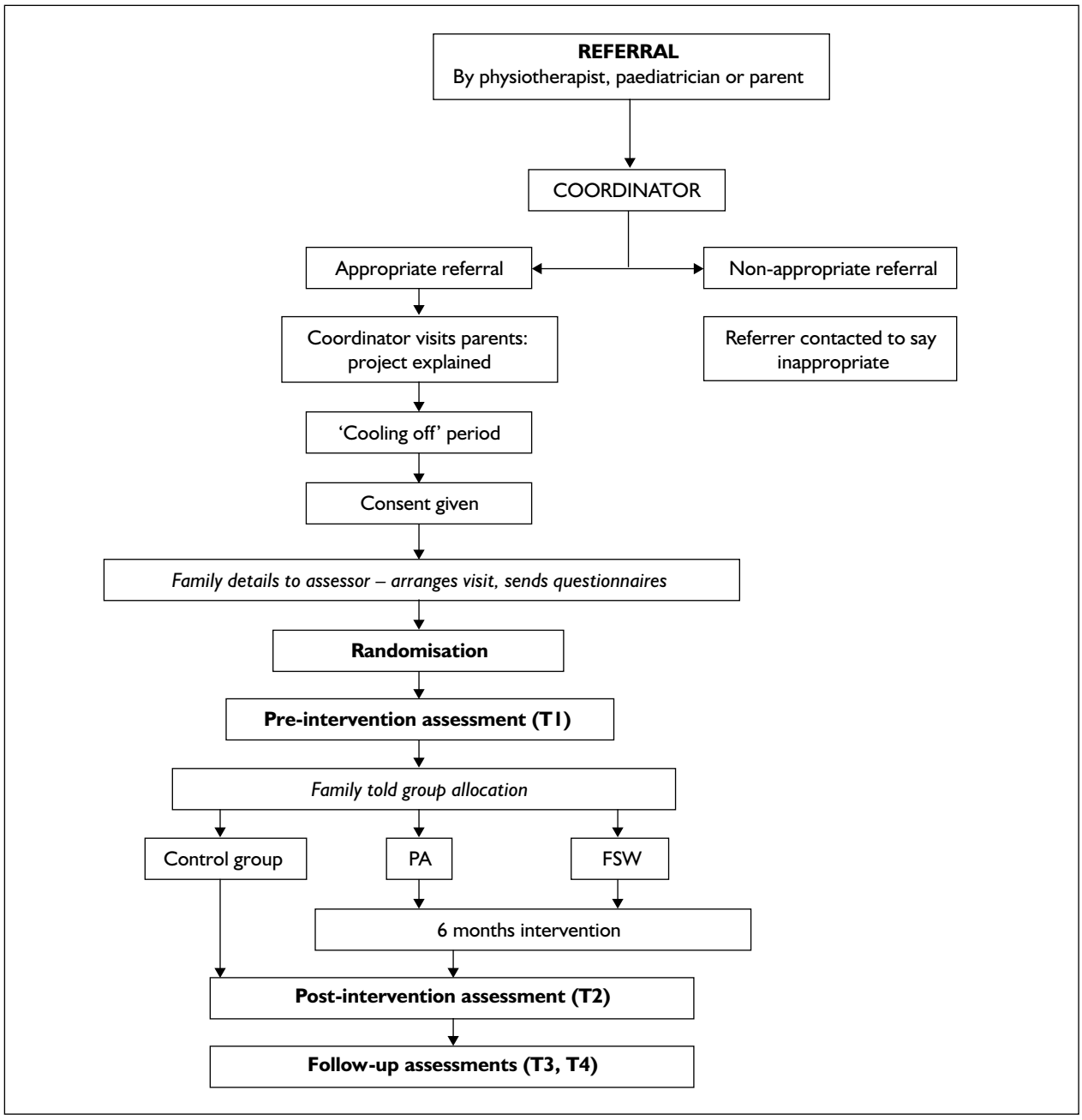

FIGURE 1 *The referral and procedure pathway*

10. The project coordinator (or independent assessor) completed the parent satisfaction interview and outlined follow-up visits.
11. Follow-up assessments were completed by the independent assessor at 6-monthly intervals (±2 weeks).
12. Newsletters about progress were sent to families who completed the intervention period at 6–12-monthly intervals.
13. A follow-up questionnaire on services received was sent out in July 2002.
14. Final feedback following the completion of the study included a newsletter to parents and a seminar for professionals.

Data quality control

Data collection

Only one data source (a follow-up questionnaire on services received) relied on postal returns. All other data were collected by the project coordinator directly from the referral agency and

then checked with the family at a home visit or by the independent assessor. The independent assessor scored the child measures and some of the family measures and gave all the completed scales and questionnaires to the project coordinator. These were checked for completion and scoring. Requests for completion were then made to the family by telephone or post.

Data entry and screening

The first 20 cases were entered into the database twice to check for accuracy. This proved to be high and was not continued to save resources. Once all the data had been entered, every third case was checked against the original scales and questionnaires. Again, the levels of accuracy were high.

Univariate descriptive statistics were used and examined for out-of-range values, plausible means and standard deviations (SDs).

Missing data were then noted and dealt with, either according to the manual for the questionnaire or, if applying to only one or two items in a scale, by imputing means. Appendix 1 describes the amount of missing data and how it was dealt with for each variable.

Each variable was checked for skewness, kurtosis and outliers. No transformations were needed as the few cases of non-normal distribution could be dealt with by bootstrapping. Further details of the methods are presented separately for each section of the results.

Reliability and validity

Appendix 1 describes all the major variables used in the study and how the reliability and validity were checked.

Reliability of the independent assessments of the GMFM and Griffiths Scale was checked by two senior paediatric physiotherapists. They accompanied the independent assessor on 15 home visits (five each at T1, T2 and T3/T4). They observed the assessment and scored the scale separately and without conferring. The agreement was over 90% in all cases.

Cronbach alpha scores were calculated when appropriate: all except one, LOC, showed reasonable to high levels of internal consistency. The patterns of correlation between the variables were also determined. All were in the expected direction and generally fell into the patterns expected from the literature (Appendix 1).

Potential problems due to biased responding to the rating scales (social desirability) were assessed using the Marlowe–Crowne Social Desirability Scale (short form)[57] and the defensive responding subscale of the PSI. The correlation coefficient was –0.42. A low score on the PSI indicates defensive responding and a high score on the Marlowe–Crowne Scale indicated socially desirable responding. Hence the two measures were associated, as expected. Since the PSI was the main parent outcome measure, the defensive subscale was used for further analysis. A score of 24 or less on the PSI subscale indicates that the defensive responding was so great that the results may be unreliable. Two mothers had a score of 24 and one a score of 21. This suggested that defensive responding and social desirability were not a cause for concern in this sample and the data obtained were reliable.

Statistical methods

Quantitative data were analysed using the statistical package STATA version 8 (StataCorp, College Station TX, USA) and the statistical tests analysis of covariance (ANCOVA), Pearson Product Moment correlations, regression analyses (including logistic regression and forwards stepwise regression) and bootstrapping were used as appropriate. (Bootstrapping is a non-parametric method which simulates data by drawing participants at random from the database and then replacing them.)

Qualitative data were analysed independently by a postdoctoral research fellow with extensive experience of qualitative data.

The diaries

The diaries kept by parents, PAs and FSWs were initially read through to provide familiarity with the nature of the text and an overview of the type of information that they contained. Particular attention was given to examining concordance between the diaries. Comments about satisfaction and additional services received were noted. A number of themes emerged, particularly from the more detailed FSW diaries. Once these themes had been exhausted, each interview was re-read and contents pertaining to the themes were noted under the appropriate heading. This procedure was followed until the contents of all of the diaries were written up within the appropriate headings. The contents within each separate heading were then read through and written up to provide a logical overview of the findings. These were then amalgamated and quotations were included where appropriate to add illustration to the text.

Chapter 3
Results

The results are reported in seven sections with a summary at the end of each section.

The participants

Referrals

Ninety families were referred from 12 CDCs. Two families withdrew soon after the initial referral. Referrals were largely from Merseyside and Cheshire. *Table 2* shows the distribution of families across the three groups according to the randomisation factors.

As can be seen from *Table 2*, 53% of referrals came from the Wirral and the neighbouring areas of Chester and Warrington, and a further 36% from the smaller areas of Widnes, Whiston and St Helen's, Ormskirk and Wigan. In these areas, an estimated 165 children would have been expected to develop CP over the study period (based on the number of births and assuming an incidence of CP of two per 1000 births; not all of these cases would have been of perinatal origin). Hence it appears that the present sample consisted of about one child in every two with CP. The groups were evenly distributed (*Table 2*).

TABLE 2 *Numbers in each group by stratification factors*

	Group			
	CG	**PAG**	**FSWG**	**Total**
N	29	28	31	88
Maternal education				
16 years	12	14	15	41
16–18 years	10	5	11	26
Higher	7	9	5	21
Pattern of spasticity				
Hemiplegia	12	11	11	34
Tetraplegia	11	11	14	36
Diplegia	6	6	6	18
Area/CDC				
Liverpool	1	1	0	2
Wirral	8	8	8	24
Chester	4	3	4	11
Warrington	4	4	4	12
Whiston/St Helen's	1	0	2	3
Ormskirk	2	2	2	6
Wigan and Leigh	3	2	3	8
Crewe	2	2	4	8
Wrexham	0	2	1	3
Widnes	2	2	2	6
Manchester	1	1	1	3
Rhyl	1	1	0	2

Withdrawals

Figure 2 shows the withdrawal and randomisation pathway.

Eighty-eight families were randomised. Before being informed of the group allocation, one child died and two withdrew at the time of the project coordinator's visit. The remaining 85 families took part in the pre-intervention assessment (T1), but one failed to complete all the self-completed questionnaires and so the family measures were only available for 84; other measures were available for all 85.

Following the assessment at T1, five more families withdrew, two before being told their group allocation and three afterwards. From comments, some parents were disappointed when they found they had not been allocated to the additional physiotherapy group. Others found the assessments excessive and intrusive. One other child died.

Seventy-nine families started the intervention period but three did not complete the intervention: two families from the FSWG (one child died and the other family was never in at the time of the home visit) and one family from the PAG when the mother became very ill.

Seventy-six families completed the intervention period and were reassessed at T2: 28 families in the CG, 25 families in the PAG and 23 families in the FSWG (*Figure 2*).

Follow-up

It was initially proposed to follow up half of the sample who completed the intervention period at 6-monthly intervals. However, because of recruitment problems, the number available for follow-up was reduced. In addition, some families were lost to follow-up because of personnel

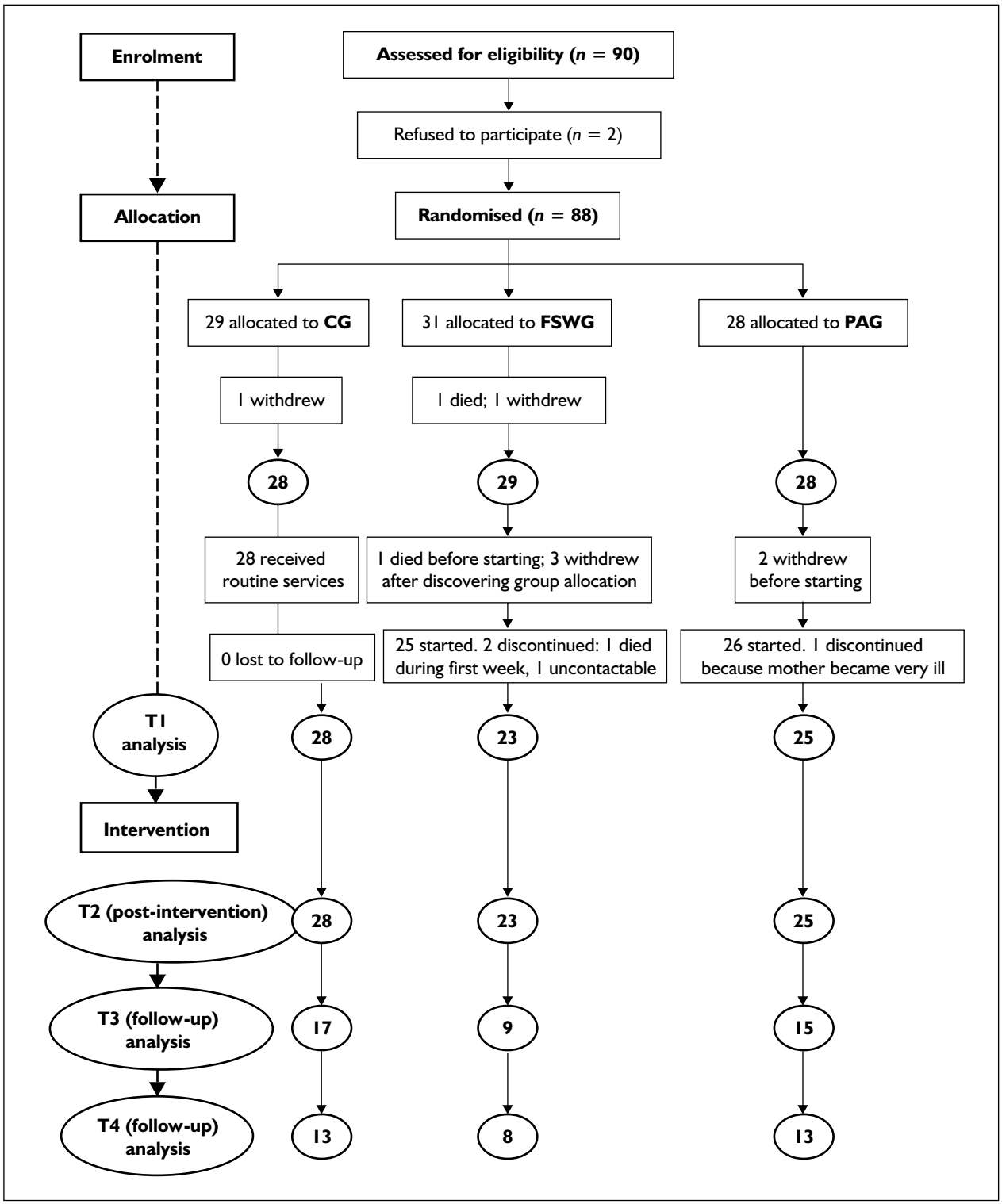

FIGURE 2 *Recruitment and participation*

changes between T2 and T3. Forty-one families (54% of the sample) were reassessed at the first follow-up (T3): there were 17 families in the CG, 15 families in the PAG and nine families in the FSWG. All families were invited for reassessment at T4 (12 months after the intervention) and 34 accepted and were assessed: 13 families in the CG, 13 in the PAG and eight in the FSWG. The power of the analyses of the longitudinal data is discussed in the appropriate sections.

Child and family characteristics

Data for all the variables are shown in *Tables 3–6*. The mean age of children entering the study was

TABLE 3 *Pre-intervention child characteristics*

Characteristic	Total group	CG	PAG	FSWG
Age of entry into study (months)				
Mean ± SD	19.8 ± 8.8	18.9 ± 8.7	19.3 ± 8.7	21.2 ± 9.2
Median	18.2	17.5	17.0	19.0
Range	9–46	9–46	8–39	8–45
Age at diagnosis (months)				
Mean ± SD	10.9 ± 6.9	10.9 ± 7.5	9.8 ± 5.9	12.1 ± 7.3
Median	8.3	8.0	8.0	11.0
Range	0–33	1–29	0–24	0–33
Gestation (weeks)				3
Mean ± SD	34.1 ± 5.3	33.2 ± 5.5	34.8 ± 4.9	4.4 ± 5.4
Median	34	33	35	33
Range	23–42	24–41	23–42	25–42
Birth weight (kg)				
Mean ± SD	2.29 ± 1.05	2.18 ± 0.99	2.43 ± 1.12	2.25 ± 1.06
Median	2.16	2.07	2.40	1.77
Range	0.60–4.7	0.74–4.7	0.60–4.6	0.88–4.0
	N **(%)**	***N*** **(%)**	***N*** **(%)**	***N*** **(%)**
Gender				
Male	52 (61)	19 (68)	16 (57)	17 (59)
Female	33 (39)	9 (32)	12 (43)	12 (41)
Natural/fostered				
Natural	83 (98)	28 (100)	27 (96)	28 (97)
Fostered	2 (2)	–	1 (4)	1 (3)
Ethnicity				
White British	79 (93)	26 (93)	26 (93)	27 (93)
Other[a]	6 (7)	2 (7)	2 (7)	2 (7)
Type of disability				
Hemiplegia	33 (39)	12 (43)	11 (39)	10 (34)
Diplegia	18 (21)	6 (21)	6 (21)	6 (21)
Tetraplegia	34 (40)	10 (36)	11 (39)	13 (45)
General health				
Well	68 (80)	20 (71)	25 (89)	23 (79)
Unwell	17 (20)	8 (29)	3 (11)	6 (21)
Birth order				
1st	39 (46)	9 (32)	15 (54)	15 (52)
2nd	32 (38)	14 (50)	8 (29)	10 (34)
3rd	6 (7)	2 (7)	1 (4)	3 (10)
4th	7 (8)	2 (7)	4 (4)	1 (3)
7th	1 (1)	1 (4)	–	–
Multiple birth				
No	75 (88)	27 (96)	25 (89)	23 (79)
Twin	7 (8)	1 (4)	–	6 (21)
Triplet	3 (4)	–	3 (11)	–

[a] 'Other' comprised 2 British-Asian, 2 Anglo-Afro-Caribbean, 1 British-Malawian and 1 British/Belgian.

19.8 months, and this was almost 9 months after the diagnosis of CP was made.

In terms of the range of measures, the children were as expected for similar samples.[58] The pattern of spastic CP was that 39% of children had hemiplegia, 40% had tetraplegia and 21% had a diplegia. Boys predominated (61%). The gestation period was shorter and the birth weight lower than normal. There were more multiple births. The mean motor and cognitive quotients fell below one SD from the norm of 100.

TABLE 4 *Pre-intervention parental characteristics*

	Total group	CG	PAG	FSWG
N	85	28	28	29
Maternal age (years)[a]				
Mean ± SD	30.9 ± 0.2	32.4 ± 3.7	30.3 ± 5.2	30.0 ± 6.3
Median	31	31	29	30
Range	17–44	26–40	18–40	17–44
Paternal age (years)[a,b]				
Mean ± SD	34.4 ± 6.6	36.1 ± 4.8	33.4 ± 4.7	33.7 ± 9.0
Median	34	35	34	33
Range	21–59	25–46	21–42	21–59
	***N* (%)**	***N* (%)**	***N* (%)**	***N* (%)**
Maternal educational qualifications				
Postgraduate	2 (2)	–	2 (7)	–
Graduate	10 (12)	3 (11)	4 (14)	3 (10)
A levels	22 (26)	10 (36)	4 (14)	8 (28)
GCSE	22 (26)	7 (25)	9 (32)	6 (21)
None	29 (34)	8 (29)	9 (32)	12 (41)
Maternal employment				
Employed	43 (51)	14 (50)	14 (50)	15 (52)
Not employed	42 (49)	14 (50)	14 (50)	14 (48)
Maternal health				
Generally well	77 (91)	26 (93)	24 (86)	27 (93)
Generally unwell	8 (9)	2 (7)	4 (14)	2 (7)
Paternal educational qualifications[b]				
Postgraduate	4 (5)	2 (7)	1 (4)	1 (3)
Graduate	11 (13)	5 (18)	2 (7)	4 (14)
A levels	17 (20)	5 (18)	6 (21)	6 (21)
GCSE	17 (20)	6 (21)	6 (21)	5 (17)
None	36 (42)	10 (36)	13 (46)	13 (45)
Paternal employment[b]				
Employed	74 (89)	25 (96)	24 (89)	25 (86)
Not employed	8 (10)	1 (4)	3 (11)	4 (14)
Paternal health[b]				
Generally well	80 (94)	26 (96)	27 (100)	27 (93)
Generally unwell	3 (4)	1 (4)	–	2 (7)

[a] One child adopted by grandparents.
[b] Data not available for some fathers.

Similarly, the family characteristics reflected those described by others.[27,28,61,62] There was a skewed distribution toward social classes 4 and 5 and lower educational levels (*Tables 4* and *5*) and the mean 'stress' scores and number of cases above threshold on the PSI and GHQ were higher than found in the normal population. The profile for PSI was similar to that reported in the manual for a sample of families with young children with CP (see Appendix 2).

Differences between the groups

Tables 3–5 present the child and family characteristics for each group. For child variables the groups were similar (*Table 3*), except for an unequal distribution of multiple births. No differences were seen for parental variables and family characteristics (*Tables 4* and *5*). For the measured variables (*Table 6*), the only variables which appeared different were (a) life stressors taken from the self-reported PSI with the FSWG reporting more and (b) COPE[47] adaptive with the FSWG having higher adaptive coping scores.

Thus, from the available knowledge of the incidence of CP and the nature of the CDCs that referred the families, there was no apparent bias

TABLE 5 *Pre-intervention custodial arrangements and family structure*

	N (%)			
	Total	**CG**	**PAG**	**FSWG**
Number of siblings				
0	32 (38)	8 (29)	14 (50)	10 (34)
1	29 (34)	14 (50)	6 (21)	9 (31)
2	14 (17)	3 (11)	3 (11)	8 (28)
3 or more	10 (11)	3 (11)	5 (18)	2 (7)
Child living with				
Both parents	66 (78)	23 (82)	19 (68)	24 (83)
Parents and grandparents	7 (8)	2 (7)	2 (7)	3 (10)
Mother only	4 (5)	1 (4)	2 (7)	1 (3)
Grandparents	1 (1)	–	1 (4)	–
Foster parents	1 (1)	–	–	1 (3)
Not stated	6 (7)	2 (7)	4 (14)	–
Primary caretaker				
Mother	75 (88)	26 (93)	24 (86)	25 (86)
Father	2 (2)	1 (4)	1 (4)	–
Mother and father	2 (2)	–	1 (4)	1 (3)
Mother, father and nanny	1 (1)	–	–	1 (3)
Grandmother	2 (2)	–	1 (4)	1 (3)
Grandmother and mother	1 (1)	–	1 (4)	–
Foster mother	1 (1)	–	–	1 (3)
Not recorded		1 (4)		
Maternal social class				
1	2 (2)	–	2 (7)	–
2	13 (15)	4 (14)	5 (18)	4 (14)
3	32 (38)	13 (46)	11 (39)	8 (28)
4	27 (31)	9 (32)	6 (21)	12 (41)
5	5 (6)	–	1 (4)	4 (14)
Not able to classify	6 (7)	2 (7)	3 (11)	1 (3)
Paternal social class				
1	3 (4)	1 (4)	2 (7)	–
2	19 (22)	8 (29)	3 (11)	8 (28)
3	29 (34)	11 (39)	11 (39)	7 (24)
4	14 (17)	1 (4)	5 (18)	8 (28)
5	17 (20)	5 (18)	6 (21)	6 (21)
Not able to classify	3 (4)	2 (7)	1 (4)	–

in the referrals. The descriptive variables and the higher levels of stress in the sample were also as expected from the available literature. Hence it appears that the sample was reasonably representative of the population of families of young children with CP in the UK.

No differences emerged between the three groups on the stratification factors. Only three variables appeared to show some difference. There were more multiple births in the PAG and FSWG groups than in the CG, with three sets of triplets in the PAG and twins in the FSWG. Self-reported Life Stressors and Adaptive Coping scores were higher in the FSWG. Since there were no differences in group means for the PSI (parent or child domain) and the GHQ, which measured actual stress experienced, this difference was not regarded as clinically significant. It was therefore concluded that the groups, despite considerable variability within them and the large number of measures, were reasonably well matched.

Short-term effects of the intervention: the main analysis

The baseline data (T1) relating to the 76 families who completed the intervention are set out in *Table 7*.

Outcome variables

Three variables were primary outcome measures: the child's GMFM, parental stress (PSI Parent

TABLE 6 *Pre-intervention baseline measures*

Measure	Total	*N*	CG	*N*	PAG	*N*	FSWG	*N*
GMFM								
Mean ± SD	35.6 ± 25.6	85	36.0 ± 27.2	28	34.6 ±23.1	28	36.3 ±27.1	29
Median	27.0		26.0		27.3		32.0	
Range	0–91.0		4.0–91.0		7.0–87.0		0–89.0	
Griffiths total score								
Mean ± SD	139.7 ± 86.6	85	144.2 ± 86.7	28	135.7 ± 72.6	28	139.3 ± 100.6	29
Median	125		123		115		129	
Range	3–446		12–313		18–326		3–446	
Vineland DL								
Mean ± SD	18.3 ± 13.1	85	17.1 ± 12.4	28	18.6 ± 9.3	28	19.2 ± 16.8	29
Median	14.0		13.5		15.5		16.0	
Range	0–73.0		0–46.0		9.0–44.0		0–73.0	
Vineland socialisation								
Mean ± SD	20.4 ± 16.6	85	20.6 ± 17.2	28	21.1 ± 15.5	28	19.5 ± 17.7	29
Median	18.0		15.0		20.0		18.0	
Range	0–56.0		0–55		0–56.0		0–54.0	
Motor impairment (MQ)								
Mean ± SD	57.6 ± 29.0	85	56.4 ± 28.6	28	61.9 ± 28.2	28	54.6 ± 30.8	29
Median	59.2		57.0		62.9		56.3	
Range	5.7–114.0		5.7–106.3		15.5–114.0		5.9–110.5	
Cognitive status (CQ)								
Mean ± SD	72.8 ± 30.0	85	73.0 ± 27.8	28	78.7 ± 29.2	28	66.8 ± 32.4	29
Median	75.9		84.8		75.8		67.0	
Range	17.1–130.3		17.1–108.5		17.5–130.3		10.0–125.0	
Social desirability								
Mean ± SD	6.8 ± 1.9	83	6.7 ± 2.0	28	7.2 ± 1.9	27	6.6 ± 1.8	28
Median	7.0		7.0		7.0		7.0	
Range	2.0–10.0		2.0–10.0		3.0–10.0		2.0–10.0	
Defensive responding (PSI)								
Mean ± SD	38.7 ± 9.7	81	38.8 ± 8.6	26	38.4 ± 10.9	27	38.8 ± 9.8	28
Median	38.0		38.5		38.0		36.0	
Range	21.0–66.0		24.0–61.0		21.0–66.0		24.0–62.0	
HOME								
Mean ± SD	40.0 ± 5.0	85	40.6 ± 4.0	28	39.7 ± 5.5	28	39.6 ± 5.5	29
Median	41.0		42.0		41.0		41.0	
Range	23.0–45.0		24.0–45.0		23.0–45.0		24.0–45.0	
COPE Adaptive								
Mean ± SD	85.5 ± 14.2	84	82.5 ± 14.6	28	82.7 ± 14.4	27	91.4 ± 12.2	28
Median	85.3		83.0		84.8		94.0	
Range	55.0–113.0		55.0–108.0		55.0–113.0		68.0–112.0	
COPE Maladaptive								
Mean ± SD	22.8 ± 5.4	84	22.9 ± 5.5	28	22.3 ± 5.5	27	23.2 ± 5.3	28
Median	22.0		22.0		22.0		22.5	
Range	14.0–39.0		15.0–37.0		14.0–35.0		16.0–39.0	
FACES Cohesion								
Mean ± SD	4.1 ± 0.6	82	4.2 ± 0.6	28	4.1 ± 0.5	26	4.1 ± 0.6	28
Median	4.1		4.2		4.1		4.2	
Range	2.3–5.0		2.3–5.0		3.1–5.0		2.4–4.8	
FACES Adaptability								
Mean ± SD	3.3 ± 0.6	82	3.3 ± 0.5	28	3.3 ± 0.6	26	3.4 ± 0.6	28
Median	3.3		3.3		3.4		3.3	
Range	2.1–5.0		2.1–4.4		2.14.8		2.1–5.0	

continued

TABLE 6 *Pre-intervention baseline measures (cont'd)*

Measure	Total	*N*	CG	*N*	PAG	*N*	FSWG	*N*
Total Family Needs								
Mean ± SD	56.1 ± 11.4	84	54.4 ± 9.4	28	56.8 ± 12.3	28	57.1 ± 12.6	28
Median	54.5		55.0		53.0		56.0	
Range	36.0–83.0		36.0–77.0		37.0–83.0		39.0–81.0	
Family support								
Mean ± SD	34.3 ± 10.1	84	34.5 ± 9.5	28	33.4 ± 11.7	28	35.0 ± 9.3	28
Median	33.0		34.0		32.0		32.0	
Range	11.0–63.0		20.0–56.0		11.0–63.0		22.0–54.0	
PSI Parent Domain								
Mean ± SD	136 ± 25	81	136 ± 22	26	138 ± 28	27	139 ± 25	28
Median	133		133		133		134	
Range	86–201		95–184		86–201		92–200	
PSI Child Domain								
Mean ± SD	116 ± 24	81	112 ± 26	26	117 ± 23	27	120 ± 24	28
Median	113		108		113		120	
Range	70–184		70–184		79–161		79–181	
GHQ								
Mean ± SD	13.4 ± 5.4	84	12.5 ± 4.9	28	13.4 ± 6.0	28	14.2 ± 5.1	28
Median	13.0		12.5		13.5		13.0	
Range	2.0–29.0		6.0–27.0		2.0–29.0		6.0–28.0	
Life Stressors								
Mean ± SD	7.1 ± 7.2	83	4.8 ± 5.0	27	6.0 ± 5.3	28	10.3 ± 9.4	28
Median	6.0		4.0		5.5		9.5	
Range	0.0–36.0		0.0–19.0		0.0–22.0		0.0–36.0	
LOC								
Mean ± SD	15.9 ± 2.9	84	15.2 ± 2.5	28	16.0 ± 2.3	28	16.5 ± 3.7	28
Median	16.0		15.0		17.0		16.0	
Range	9.0–26.0		9.0–20.0		12.0–20.0		10.0–26.0	

TABLE 7 *Pre-intervention baseline measures on 76 children and families who completed the intervention (T1)*

	Total		CG		PAG		FSWG	
Measure	Mean ± SD	*N*	Mean ± SD	*N*	Mean ± SD	*N*	Mean ± SD	*N*
GMFM	36.7 ± 25.3	76	36.0 ± 27.2	28	35.6 ± 23.3	25	38.9 ± 25.9	23
Griffiths total score	141.2 ± 81.2	76	144.2 ± 86.7	28	136.2 ± 76.6	25	143.0 ± 82.6	23
Vineland DL	18.1 ± 12.1	76	17.1 ± 12.4	28	19.0 ± 9.7	25	18.3 ± 14.3	23
Vineland socialisation	20.8 ± 16.8	76	20.6 ± 17.2	28	21.2 ± 16.4	25	20.4 ± 17.3	23
Motor quotient (MQ)	59.7 ± 28.6	76	56.4 ± 28.6	28	63.6 ± 28.5	25	59.4 ± 29.3	23
Cognitive quotient (CQ)	74.7 ± 29.6	76	73.0 ± 27.8	28	79.5 ± 30.8	25	71.3 ± 30.8	23
Social desirability	6.9 ± 1.8	76	6.7 ± 2.0	28	7.2 ± 1.9	25	6.7 ± 1.5	23
Defensive responding (PSI)	37.8 ± 9.4	74	38.8 ± 8.6	26	38.0 ± 11.2	25	36.6 ± 8.3	23
HOME	40.3 ± 4.5	76	40.6 ± 4.0	28	40.2 ± 4.6	25	40.0 ± 5.1	23
COPE Adaptive	85.0 ± 14.6	76	82.5 ± 14.6	28	82.4 ± 15.3	25	90.8 ± 12.6	23
COPE Maladaptive	22.6 ± 5.2	76	22.9 ± 5.5	28	22.4 ± 5.7	25	22.5 ± 4.4	23
Mean FACES III Cohesion	4.1 ± 0.54	76	4.2 ± 0.62	28	4.1 ± 0.47	25	4.1 ± 0.50	23
Mean FACES III Adaptability	3.3 ± 0.56	76	3.3 ± 0.53	28	3.3 ± 0.53	25	3.4 ± 0.63	23
Total Family Needs	55.0 ± 11.0	76	54.4 ± 9.4	28	56.5 ± 12.5	25	54.0 ± 11.2	23
Family support	34.4 ± 10.1	76	34.5 ± 9.5	28	34.3 ± 12.0	25	34.4 ± 8.7	23
PSI Parent Domain	135.6 ± 24.2	74	135.7 ± 21.7	26	136.9 ± 28.5	25	133.9 ± 22.7	23
PSI Child Domain	115.2 ± 23.4	74	111.8 ± 25.5	26	116.1 ± 23.3	25	118.0 ± 21.6	23
GHQ	13.0 ± 5.2	76	12.5 ± 4.9	28	13.4 ± 6.4	25	13.1 ± 4.4	23
Life Stressors	6.6 ± 6.6	75	4.8 ± 5.0	27	6.1 ± 5.5	25	9.3 ± 8.4	23
LOC	15.7 ± 2.7	76	15.2 ± 2.5	28	16.0 ± 2.4	25	16.0 ± 3.3	23

TABLE 8 *Outcome variables at T2*

	CG		PAG		FSWG	
	N	**Mean ± SD**	***N***	**Mean ± SD**	***N***	**Mean ± SD**
Child outcomes at T2						
GMFM	28	45.5 ± 29.7	25	50.0 ± 25.8	23	48.0 ± 30.7
Vineland DL	28	24.5 ± 17.1	25	25.5 ± 11.0	22	25.5 ± 16.3
Griffiths raw score	28	185.6 ± 114.9	25	188.8 ± 98.7	23	185.7 ± 106.8
Family outcomes at T2						
PSI (Parent Domain)	27	134.1 ± 25.3	25	140.1 ± 33.3	22	136.5 ± 22.2
Family Needs	28	54.4 ± 8.1	25	56.8 ± 13.3	23	55.6 ± 14.7

Domain) and Family Needs. Two secondary measures examined the effect of the intervention on the child's wider functioning. These were the Vineland DL and Griffiths scales (the Griffiths total raw score was used as it gives a measure of general developmental attainment). The Vineland DL correlated with the Socialisation score ($r = 0.88$), and was selected as it was more likely to reflect abilities in motor gains.

Table 8 gives means and SDs for each outcome variable at T2, broken down by experimental group.

Statistical methods

The analysis of the RCT was guided by the recommendations of the European Agency for the Evaluation of Medicinal Products (EMEA).[63,64]

The study aimed to investigate whether gains in child outcomes would be greater for families receiving an intervention by a PA than for the CG, and whether gains in family outcomes would be greater for the FSWG than for the CG. The group variable was entered into each analysis as two 'dummy' or indicator variables so as to partition the sums of squares into two components, the first comparing the PAG with the controls and the second comparing the FSWG with the controls. Only the components relevant to each hypothesis are reported (i.e. component 1 for child outcomes and component 2 for family outcomes).

Covariates

The covariates used in the primary analysis were the baseline values of the outcome measure and the factors used to stratify the sample for randomisation: pattern of spasticity (three levels: hemiplegia, tetraplegia, diplegia); the mother's education (three levels: up to age 16, age 16–18, age over 18 years (higher education)); and CDC (12 levels, collapsed to two; see below).

The large number of CDCs relative to sample size, coupled with the small numbers recruited from most (*Table 1*), meant that the factor could not be used as a covariate in its original form. A much larger number of families (24) were recruited from one CDC, Wirral, than from any other. Furthermore, there were a number of important clinical differences between the Wirral CDC and the other centres. In particular, on the Wirral, families received a higher level of service support: disclosure of the diagnosis of CP tended to be at a younger age, and the CDC was particularly keen to support the study [see the section 'Multivariate analysis' (p. 33) for details]. There were no notable clinical differences between the other CDCs and therefore for analysis purposes we collapsed CDC to two levels, Wirral versus non-Wirral.

Primary analyses

The analyses were conducted on an intention-to-treat basis, using ANCOVA with robust estimates of variance. All analyses were undertaken using STATA version 8. Each outcome was subjected to a series of ANCOVA analyses. The primary analysis used the baseline values of the outcome and the stratification factors as covariates, as indicated above. Bootstrapping was used to cross-check the significance or otherwise of p-values falling below 0.2 because of non-normality in the distributions of the outcomes. To assess the robustness of the results from the primary analysis, two subsequent sensitivity analyses were conducted.

Sensitivity analysis

Withdrawal from the study after randomisation but before data collection at T2 varied between groups: one (3%) family withdrew from the CG, three (11%) families from the PAG, and eight (26%) families from the FSWG. Stepwise logistic regression was applied to determine if any child or family variables (at baseline T1) predicted withdrawal. The only variable to enter the equation at a significance level of $\alpha = 0.05$ was

TABLE 9 *Summary of ANCOVA analyses of child outcomes at T2*

	PAG vs CG				
	F	***df***	***p***	**Estimated effect size (95% CI)**	***p* using bootstrap method[a]**
GMFM (primary outcome)					
Including covariates[b]	3.31	1, 67	0.07	5.0 (–0.5 to 10.4)	0.04
Including covariates and weights[c]	3.39	1, 67	0.07	4.9 (–0.4 to 10.3)	0.04
Unadjusted analysis[d]	2.76	1, 72	0.10	5.0 (–1.0 to 11.1)	0.09
Vineland DL (secondary outcome)					
Including covariates	0.30	1, 66	0.58	–1.0 (–4.6 to 2.6)	–
Including covariates and weights	0.18	1, 66	0.67	–0.8 (–4.4 to 2.6)	–
Unadjusted analysis	0.45	1, 71	0.49	–1.4 (–5.3 to 2.6)	–
Griffiths raw score (secondary outcome)					
Including covariates	0.41	1, 67	0.53	5.3 (–11.3 to 21.8)	–
Including covariates and weights	0.27	1, 67	0.61	4.1 (–11.9 to 20.2)	–
Unadjusted analysis	0.44	1, 72	0.51	5.4 (–10.9 to 21.7)	–

[a] Bootstrap was only applied if the ANCOVA gave $p < 0.2$.
[b] ANCOVA model including group, outcome at T1 and stratification factors.
[c] Weights related to the probability of withdrawal from the study added to the model.
[d] ANCOVA model including group and outcome at T1 only.

Family Needs. The results of the logistic analysis were used to estimate the probability of participation at T2 on the basis of the Family Needs score, and the inverse of these probabilities was then assigned to individual cases for use as probability weights in a sensitivity analysis. This method provides some adjustment for baseline imbalance that may have resulted from differential withdrawal and for cases that are missing at random (i.e. where the reason for withdrawal is independent of the missing values). However, it does not adjust for withdrawal related to outcome,[65] which may well apply in the present study. A number of methods have been proposed for this situation, but there is no universally accepted methodology and different approaches can lead to different results,[64] therefore these methods have not been applied here.

A second sensitivity analysis was conducted to check the robustness of the findings to the choice of covariates.[63] The use of the collapsed CDC variable as a covariate does not fully reflect the restriction on randomisation implied by the original stratification, and other covariates were not prespecified but based on stratification factors. In view of this, we used the safest (most conservative) option[63] and did a sensitivity analysis unadjusted for any covariates (other than the baseline values of the outcome measure) or for the probability weights. It has been proposed that an unadjusted analysis should always be presented alongside the adjusted analysis.[66]

Any important differences between the results of the sensitivity analyses and the primary analysis are discussed.

Results for the child outcomes

Table 9 summarises results from the ANCOVA analyses comparing the PAG with the CG on the three child outcomes.

Gross Motor Function Measure (primary outcome)

The change in GMFM between the assessments at T1 and T2 was similar for the three groups.

The primary analysis comparing GMFM between the children in the PAG and those in the CG at T2, controlling for outcome scores and covariates at T1, was not statistically significant using the parametric test ($p = 0.07$), but was significant using bootstrapping ($p = 0.04$). In the first sensitivity analysis, when probability weights were added to the model, identical results were obtained. However, the second sensitivity analysis, in which no covariates or adjustments were applied, did not reach significance by either the parametric ($p = 0.1$) or bootstrapped ($p = 0.09$) tests.

Vineland Daily Living and Griffiths raw score (secondary outcomes)

There was no significant difference between the PAG and the CG for both of the primary analysis and the two sensitivity analyses for both these variables (*Table 9*).

TABLE 10 *Summary of ANCOVA analyses of family outcomes at T2*

	FSWG vs CG				
	F	***df***	***p***	**Estimated effect size (95% CI)**	***p* using bootstrap method[a]**
PSI (Parent Domain) (primary outcome)					
Including covariates[b]	0.30	1, 65	0.58	2.7 (–7.1 to 12.5)	–
Including covariates and weights[c]	0.14	1, 65	0.71	1.8 (–8.0 to 11.5)	–
Unadjusted analysis[d]	0.61	1, 70	0.44	3.9 (–6.1 to 14.0)	–
Family Needs (primary outcome)					
Including covariates	0.04	1, 67	0.83	0.7 (–4.7 to 5.9)	–
Including covariates and weights	0.04	1, 67	0.85	0.5 (–4.9 to 5.9)	–
Unadjusted analysis	0.25	1, 72	0.62	1.4 (–4.3 to 7.2)	–

[a] Bootstrap was only applied if the ANCOVA gave $p < 0.2$.
[b] ANCOVA model including group, outcome at T1 and stratification factors.
[c] Weights related to the probability of withdrawal from the study added to the model.
[d] ANCOVA model including group and outcome at T1 only.

Results for the family outcomes

PSI Parent Domain and Family Needs (primary outcomes)

There was no significant difference between the FSWG and the CG for both of these variables (*Table 10*).

Interim discussion and conclusions

No evidence was found to support the three primary hypotheses. In particular, there was no difference using ANCOVA. However, when bootstrapping was used, the increase in GMFM just reached significance. This gave us considerable concern. In order to check whether a true difference had been missed, the results were cross-referenced with two other measures of motor development, the raw score of the Griffiths locomotor subscale, and the MQ. Both of these measures correlated closely with the GMFM at around the $r = 0.70$ level. Both clearly failed to reach significance (locomotor score, $p = 0.94$; MQ, $p = 0.79$). Considering the 95% CIs around the estimated effect size, the highest possible increase in GMFM was 10.4 points. Even this was almost four points less than the clinically significant level of change (14 points) that had been prespecified at the planning stages of this study. Finally, we considered whether a Type 2 error might have occurred and the arguments against this possibility are presented in the final discussion.

The effect of differential drop-out was also considered. Withdrawal from the study was fairly minimal and not too dissimilar for the controls and PA group. Because there were more drop-outs from the FSWG and the main predictor of drop-out was a higher Family Needs score, differential drop-out was only pertinent to parental outcomes and the comparison between the FSWG and CG. Families that withdrew from the FSWG prior to T2 had a mean Family Needs score at baseline of 66.8 compared with 55.0 for families that participated at T2. Their withdrawal was therefore likely to have lowered the mean Family Needs score for the FSWG at T2, compared with if they had participated at T2. Hence withdrawal would have increased the group difference at T2, making it more (rather than less) likely for an effect of the intervention to be detected if it existed.

Medium-term effects: analysis of the follow-up data

Although no intervention effects were seen at T2, the possibility of later effects was examined. The data at T3 and T4 were subjected to the same form of ANCOVA as used above. The reduced size of groups at T3 and T4 decreased the power of the analyses and therefore only the unadjusted analysis was performed.

Table 11 presents the means and SDs obtained by the three groups at T3 and T4 for the three child outcome measures and the two family outcome measures.

Results for child outcomes

Gross Motor Function Measure (primary outcome)

The ANCOVA analyses of GMFM at T3 and T4 did not find any significant differences between

TABLE 11 *Outcome variables at T3 and T4*

	CG		PAG		FSWG	
	N	**Mean ± SD**	***N***	**Mean ± SD**	***N***	**Mean ± SD**
Child outcomes						
GMFM at T3	17	55.7 ± 32.9	15	61.5 ± 27.4	9	51.7 ± 28.6
GMFM at T4	13	64.5 ± 32.5	13	64.2 ± 31.6	8	62.5 ± 24.8
Vineland DL at T3	17	31.4 ± 15.2	15	27.7 ± 12.8	9	31.7 ± 13.5
Vineland DL at T4	10	34.8 ± 15.6	11	33.9 ± 15.1	8	41.1 ± 12.0
Griffiths at T3	17	204.5 ± 82.8	15	192.3 ± 79.9	9	195.0 ± 75.6
Griffiths at T4	13	228.5 ± 85.6	13	216.1 ± 87.6	8	238.9 ± 54.8
Family outcomes						
PSI at T3	15	126.5 ± 16.9	15	143.7 ± 38.0	8	142.5 ± 14.9
PSI at T4	12	131.3 ± 14.4	11	143.8 ± 32.3	8	137.4 ± 22.4
Family Needs at T3	16	52.5 ± 8.8	15	55.0 ± 13.2	8	47.0 ± 5.7
Family Needs at T4	11	55.3 ± 6.5	11	48.6 ± 12.5	7	42.9 ± 4.8

TABLE 12 *Summary of analyses of longitudinal outcomes for GMFM*

	PAG vs CG				
	F	***df***	***p***	**Estimated effect size (95% CI)**	***p* using bootstrap method[a]**
GMFM at T3					
Unadjusted analysis[b]	1.64	1, 37	0.21	8.9 (–5.2 to 23.0)	–
GMFM at T4					
Unadjusted analysis	0.14	1, 30	0.71	3.4 (–15.3 to 22.0)	–

[a] Bootstrap was only applied if the ANCOVA gave $p < 0.2$.
[b] Model including group and GMFM at baseline as independent variables.

the PAG and the CG (*Table 12*). *Figure 3* shows the GMFM scores for these children at each time point.

Vineland Daily Living and Griffiths raw score (secondary outcomes)

None of the analyses at separate time points reached statistical significance.

Results for family outcomes

PSI Parent Domain (primary)

The analyses of T3 and T4 data did not show any statistically significant differences in stress levels between parents who received family worker support and those in the CG.

Family Needs (primary)

The results are presented in *Table 13* and *Figure 4*. A significant difference was found between groups at T4 ($p = 0.001$), but not at T3 ($p = 0.33$).

At T4, the FSWG had an average Family Needs score that was lower than that for the CG by 12 points (95% CI –18 to –6). The average difference of 12 points represents a 22% reduction in mean Family Needs score (or, for example, a reduction from six items being a definite need to no need, or a reduction from 12 items being a possible need to no need).

Effect of withdrawals on the longitudinal data

The longitudinal analysis found a significant effect for Family Needs at T4. *Figure 5* presents graphs of mean Family Needs scores by follow-up time, broken down by time of withdrawal and group.

Families in the FSWG, who withdrew following T1 or T2, had considerably higher mean scores prior to withdrawal compared with families in the FSWG who participated to the end of the study. FSWG families that participated throughout showed a consistently decreasing mean Family Needs score over time, suggesting that only those families whose needs were being progressively met

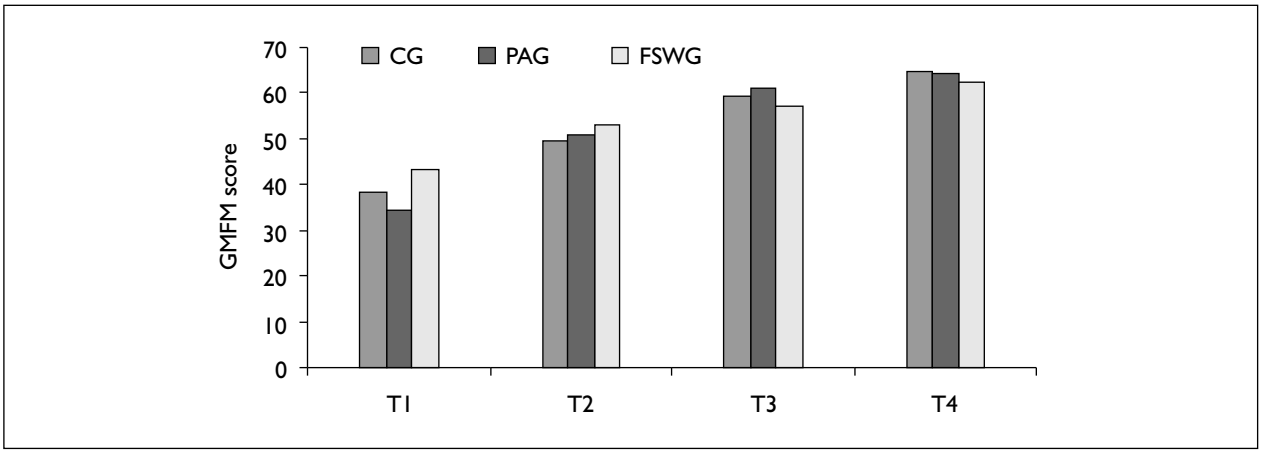

FIGURE 3 *The mean gains in GMFM scores from T1 to T4 for the 34 children who were in the study at all four time points. There were 13 children in the CG, 13 in the PAG and eight in the FSWG.*

TABLE 13 *Summary of longitudinal analyses of family outcomes*

	FSWG vs CG				
	F	***df***	***p***	**Estimated effect size (95% CI)**	***p* using bootstrap method[a]**
PSI (primary outcome) at T3 Unadjusted analysis[b]	1.25	1,34	0.27	8.0 (–6.6 to 22.7)	–
PSI (primary outcome) at T4 Unadjusted analysis	0.004	1,27	0.95	–0.4 (–14.8 to 13.9)	–
Family Needs (primary outcome) at T3 Unadjusted analysis	0.98	1,35	0.33	–3.9 (–11.9 to 4.1)	–
Family Needs (primary outcome) at T4 Unadjusted analysis	14.75	1,25	0.001	–12.0 (–18.4 to –5.6)	<0.001

[a] Bootstrap was only applied if the ANCOVA gave $p < 0.2$.
[b] Model including group and outcome at baseline as independent variables.

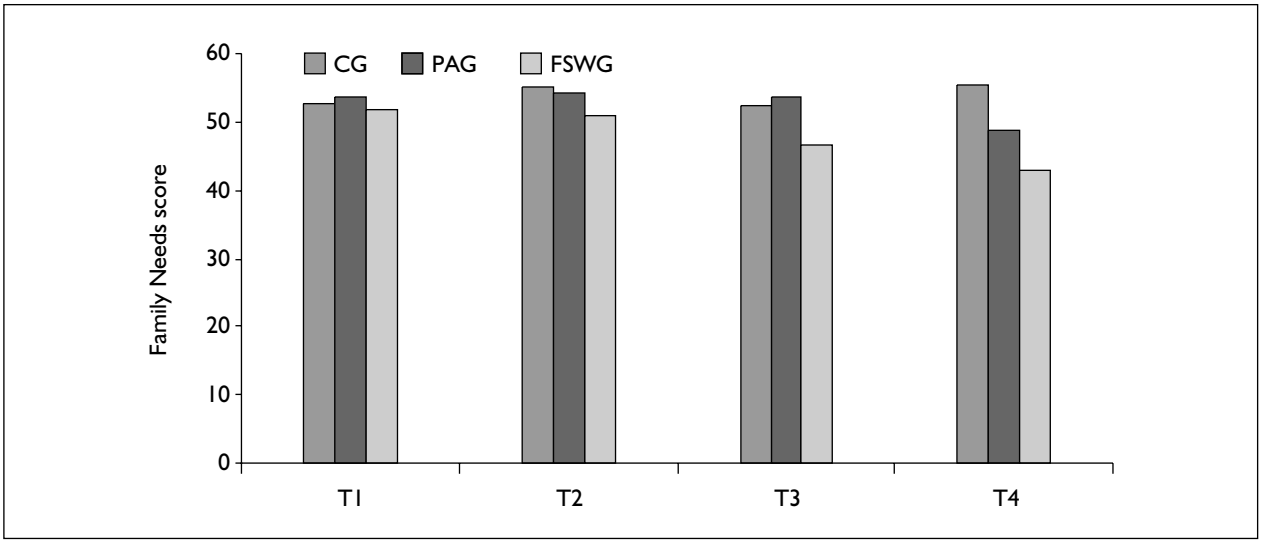

FIGURE 4 *The mean Family Needs scores of infants in the study for the four assessments. There were 11 children in the CG, 11 in the PAG and seven in the FSWG.*

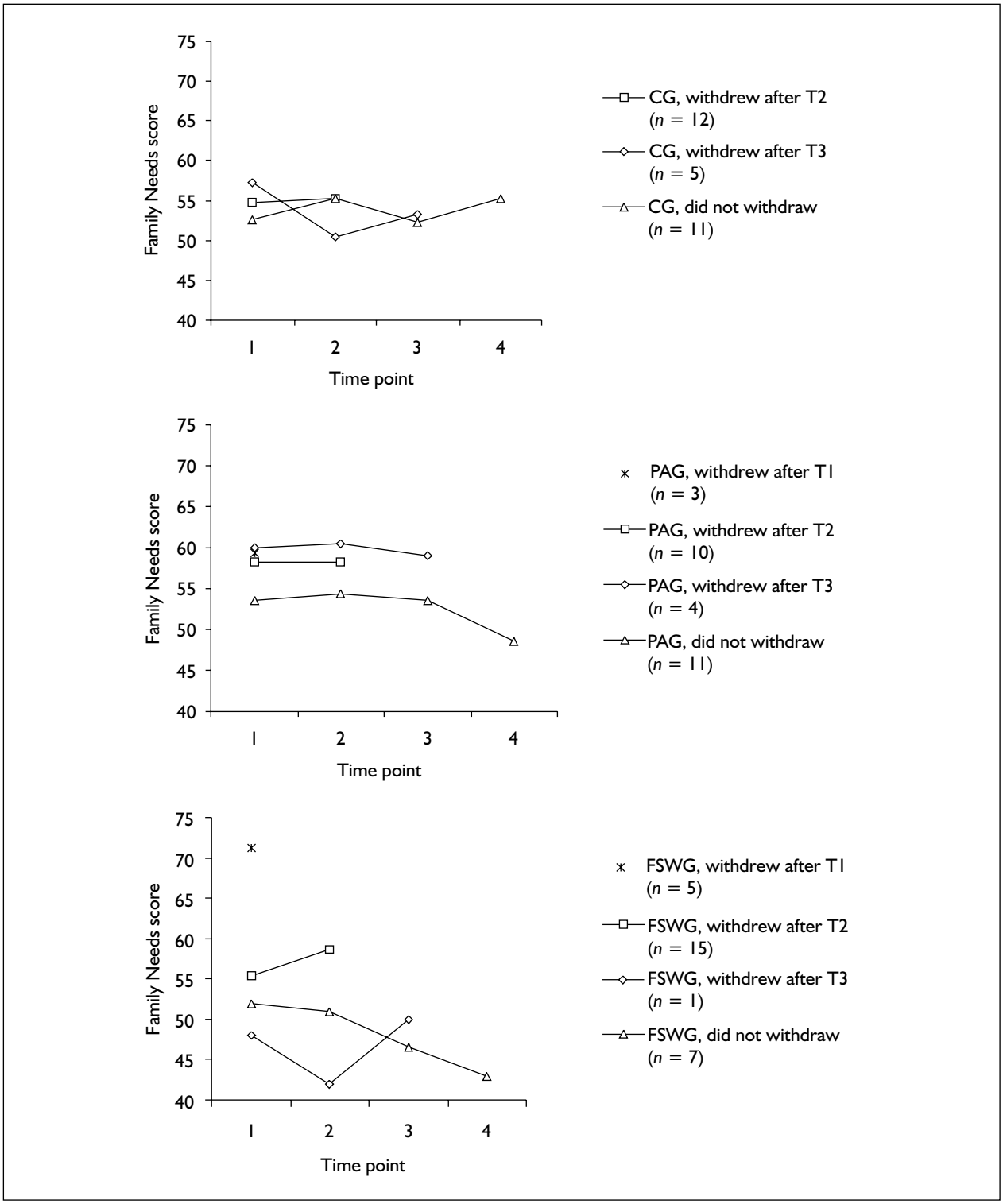

FIGURE 5 *Mean Family Needs score by time of withdrawal from study for the CG, PAG and FSWG*

continued to participate. In contrast, mean Family Needs scores for CG families showed no trend over time and appeared unrelated to withdrawal. The PAG appeared to be similar to the FSWG in that withdrawals tended to be families with higher Family Needs scores. The lack of any trend over time within the CG suggested that it would be reasonable to apply the last-observation-carried-forward method to explore visually trends within the complete groups (i.e. including withdrawals). These plots (*Figure 6*) show little evidence of any substantive differences between the groups.

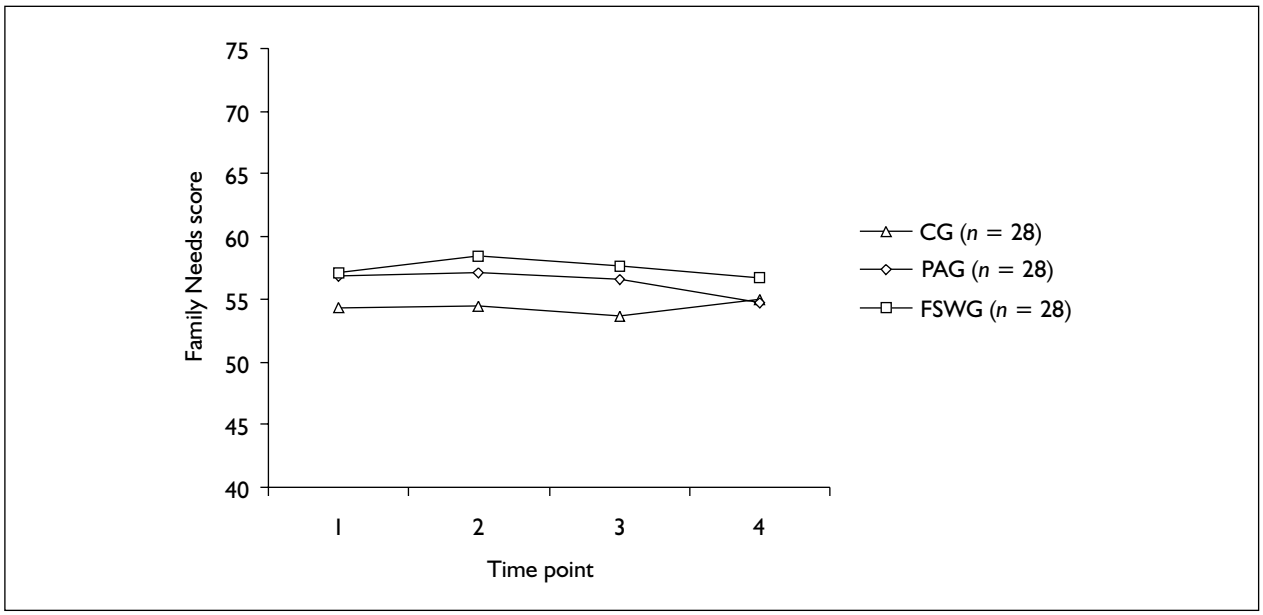

FIGURE 6 *Mean Family Needs scores by group, using last-observation-carried-forward*

Interim discussion and conclusions

The reduced numbers of families at T3 and T4 meant that the power of the study to detect a difference at these time points was limited. There were no significant differences for any of the child outcome measures. For parental outcomes, the FSWG had significantly lower Family Needs scores at T4.

Interpretation of these results was complicated by the level of withdrawal from the longitudinal aspect of the study, and the fact that withdrawal was commonly associated with higher Family Needs scores, most notably amongst the FSWG.

It appeared that the significantly lower FSWG Family Needs score at T4 was principally due to the continuing participation of a subgroup of FSW families whose family needs were being progressively met.

Cost-effectiveness analysis

The primary purpose of incorporating economic evaluation alongside the trial of additional physiotherapy for children with CP was to identify whether additional intervention by a PA was cost-effective in terms of children's gross motor function, as compared with usual levels of physiotherapy, or the impact and cost of the family receiving help from an FSW. Such costs need to be evaluated against those of routine services. Hence levels of current service receipt and their costs were examined. As noted in Chapter 1, there was little information about service input and costs for young children with CP.

Contacts with services

Staff and families were asked to record the frequency and type of contacts that they had with a wide range of healthcare, social care and educational and voluntary services over the 6-month intervention period of the trial (see Appendix 1). *Table 14* shows the range of services with which families had contact and the frequency of those contacts. Families were included only when there were sufficient data for the cost analysis. *Figure 7* shows the mean number of service contacts per child for each group in graphical form.

Unit costs of services at 2002 prices

Table 15 shows the unit costs of services at 2002 prices. These were national reference costs published by the Personal Social Services Research Unit, University of Kent, Canterbury,[67] by the NHS (2002) and by the National Union of Teachers (online). Where exact costs could not be sourced for a particular service, similar services were chosen as proxies and relevant details are given in *Table 15*; for example, in the case of the O_2 nurse, no specific reference cost could be sourced, so an HIV/AIDS nurse cost was used on the assumption that a similar level of specialist training would be involved.

Where unit costs were obtained from Netten and Curtis,[67] a general choice was made to include

TABLE 14 *Frequency of service contacts*

Service	Control group N^a = 28		Intervention group N^a = 25		FSW group N^a = 21	
	Mean no. of contacts per child	SD	Mean no. of contacts per child	SD	Mean no. of contacts per child	SD
Visits to GP surgery	3.29	4.05	2.04	1.37	4.29	5.91
Domiciliary visits by GP	0.32	0.77	0	0	0.19	0.51
Hospital outpatient	4.18	3.04	3.72	2.28	5.43	2.73
Hospital inpatient	0.39	0.69	0.36	0.86	1.10	2.77
Attend A&E	0.11	0.32	0.28	0.54	0.24	0.44
Physical therapy session	32.11	27.26	34.60	23.41	32.29	24.45
Occupational therapist	9.50	10.27	9.36	9.91	10.24	14.68
Community nurse	1.57	1.64	2.32	5.15	1.86	2.80
Speech therapist	5.29	8.63	3.52	5.84	4.48	8.35
Audiologist	0.25	0.65	0.08	0.40	0	0
Portage	4.64	10.14	5.20	10.61	6.19	11.35
Ophthalmologist	0	0	0.08	0.28	0	0
Orthoptist	0.46	0.69	0.24	1.01	0.14	0.66
Orthotist	0.75	1.51	1.40	2.02	0.71	1.10
Dietician	0.21	0.69	0.40	0.82	0.52	1.54
Dentist	0.04	0.19	0	0	0.05	0.22
Teacher for deaf	0.89	3.86	0	0	0	0
O_2 nurse	0	0	0	0	0.24	1.09
Nursery officers	0	0	0	0	0.95	4.36
Teacher for visually impaired	0.04	0.19	0	0	0	0
Play therapist	0	0	0	0	1.33	6.11
FSW	0	0	0	0	18.00	0
PA	0	0	18.00	0	0	0

[a] *N* = number of children in group.

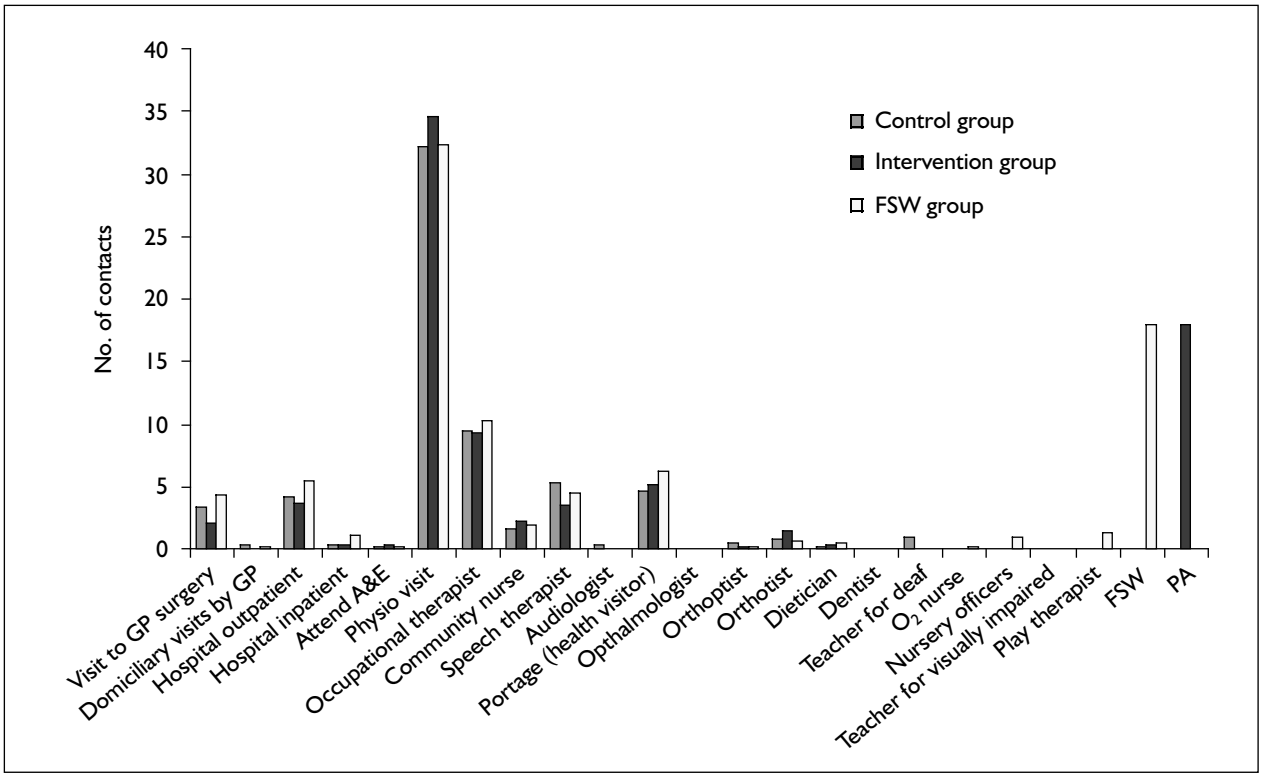

FIGURE 7 *Mean number of contacts per child by type of service. 'Physio visit' denotes physiotherapy sessions. This includes all contacts with physiotherapists and also portage, visits to CDCs and toy libraries and hydrotherapy.*

TABLE 15 *Unit costs of services*

Service	Unit cost (£)	Unit	Source of unit cost information[a]
Visits to GP surgery	20	Surgery consultation	Netten and Curtis, 2002: 116
Domiciliary visits by GP	61	Visit	Netten and Curtis, 2002: 116
Hospital outpatient	111	Per paediatrics outpatient day	Netten and Curtis, 2002: 95
Hospital inpatient	398	Per paediatrics inpatient day	Netten and Curtis, 2002: 95
Attend A&E	57	Per day hospital attendance	Netten and Curtis, 2002: 95
Physical therapy session	26	Per hour	Netten and Curtis, 2002: 101 (community physiotherapist cost used)
Occupational therapist	26	Per hour	Netten and Curtis, 2002: 146
Community nurse	26	Per hour	Netten and Curtis, 2002: 109
Speech therapist	25	Per hour	Netten and Curtis, 2002: 147
Audiologist	50	Per contact	National Reference Costs, 2002: 161–2.
Portage (nursery nurse)	10	Per hour	Netten and Curtis, 2002: 113 (community auxiliary nurse cost used as a proxy)
Ophthalmologist	90	Per contact	Netten and Curtis, 2002: 164 (cost for medical consultant used)
Orthoptist	26	Per hour	Netten and Curtis, 2002 (physiotherapist/occupational therapist cost used as proxy)
Orthotist	26	Per hour	Netten and Curtis, 2002 (physiotherapist/occupational therapist cost used as proxy)
Dietician	25	Per hour	Netten and Curtis, 2002: 148
Dentist	42.33	Per visit	National Reference Costs, 2002: 180, 182, 184
Teacher for deaf	26	Per hour	Based on upper scale 2 grade (National Union of Teachers; online)
O_2 nurse	27	Per hour	Netten and Curtis, 2002: 112 (NHS community nurse specialist for HIV/AIDS used as proxy)
Nursery officers	26	Per place per session	Netten and Curtis, 2002: 88
Teacher for visually impaired	26	Per hour	Based on upper scale 2 grade (National Union of Teachers; online)
Play therapist (portage)	10	Per hour	Netten and Curtis 2002 (physiotherapist/occupational therapist cost used as proxy)
FSW	17	Per hour	Netten and Curtis, 2002: 130
PA	10	Per hour	Netten and Curtis, 2002: 113 (community auxiliary nurse cost used as a proxy)

[a] Netten and Curtis, 2002 = Ref. 69.

qualification costs where available. Where separate costs per client contact hours, per home-visit hours, per clinic hour and per standard hours were available, standard hourly costs were used which incorporate costs for all activity types.

Although some costs were given in units of visits or attendances (e.g. GP consultations), most costs were given per hour. Because participants in the study were asked about the number of contacts with professionals and services, not the number of

TABLE 16 *Mean cost per child for overall services plus additional services*

Group	*N*	Mean cost per child ± SD (£)
Control	28	2087.90 ± 1297.88
PA		252200.08 ± 956.20
FSW	21	2865.02 ± 1306.91

contact hours, this led to assumptions being made about the duration of contacts. In most cases, we estimated that each contact entailed 1 hour of professional's time (e.g. speech therapist, occupational therapist, Portage Home Visiting service).

Costs for teachers of the deaf and teachers of the blind were determined by personal communication with the Secretary of the British Association of Teachers of the Deaf, who advised that teachers of the deaf and blind receive standard teaching salaries plus two increments. Therefore, we estimated the cost of these teachers' time by taking the middle point of the main and upper teaching salary scales (main scale 6) and adding two incremental spine points (to upper scale 2). An hourly cost was then estimated by comparing these salary figures with similar published health service unit costs.

As the intervention period was of only 6 months' duration, no discount rate was applied to costs.[68]

Assumptions

The analysis made the following assumptions regarding length and frequency of service contacts with physiotherapy assistants, family support workers and Portage:

1. PAs were estimated to have delivered a total of 18 hours to children in the PAG group over the 6-month study period.
2. Similarly, FSWs were also estimated to have delivered 18 hours over the 6-month study period to children in the FSWG.
3. Children from all three treatment groups who received Portage were estimated to have received 26 service hours of home visits over the 6-month study period.

Total mean cost per child

Combining the data on frequency of service contact with the unit costs of services provided a total cost for each child in the study, which can be averaged to supply mean costs per child for each arm of the trial, as shown in *Table 16*. The histograms in *Figures 8–10* show the distributions and normal curves for total cost for each child in each of the three arms of the trial.

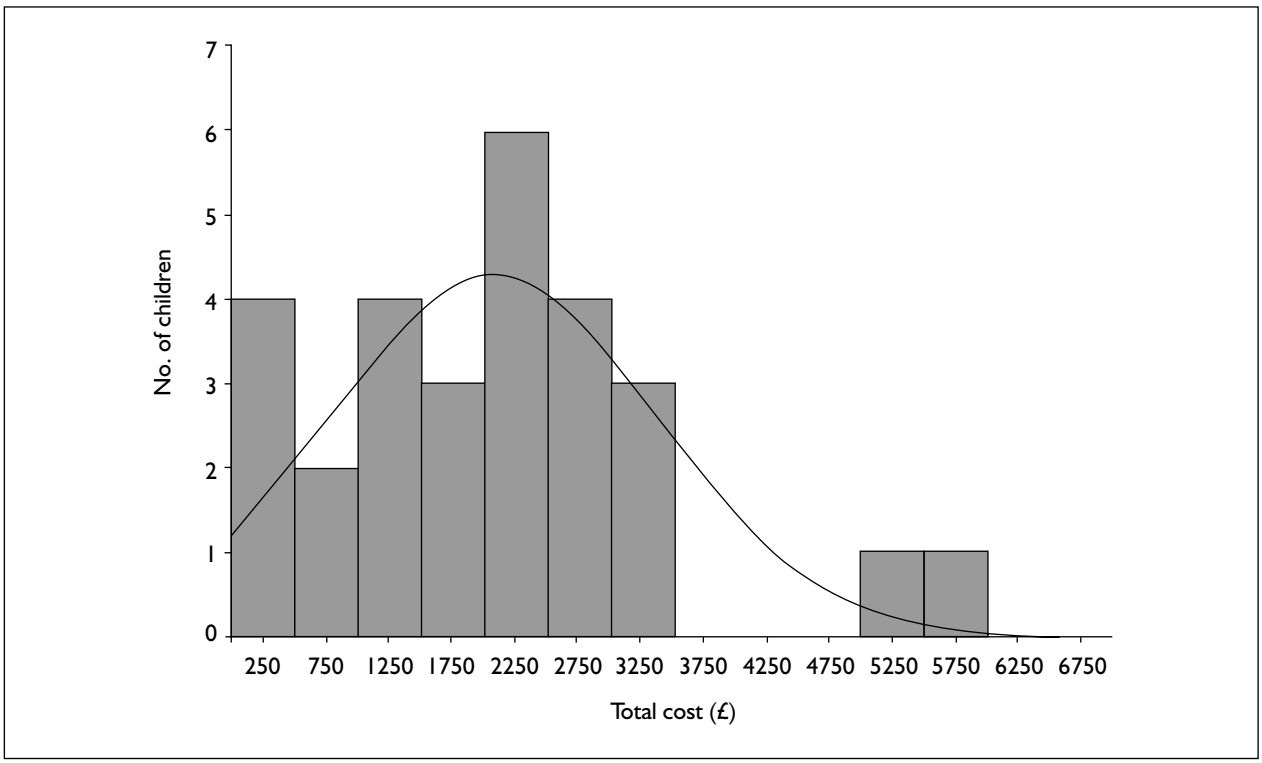

FIGURE 8 *Distribution and normal curve of total cost for each child in the control arm of the trial*

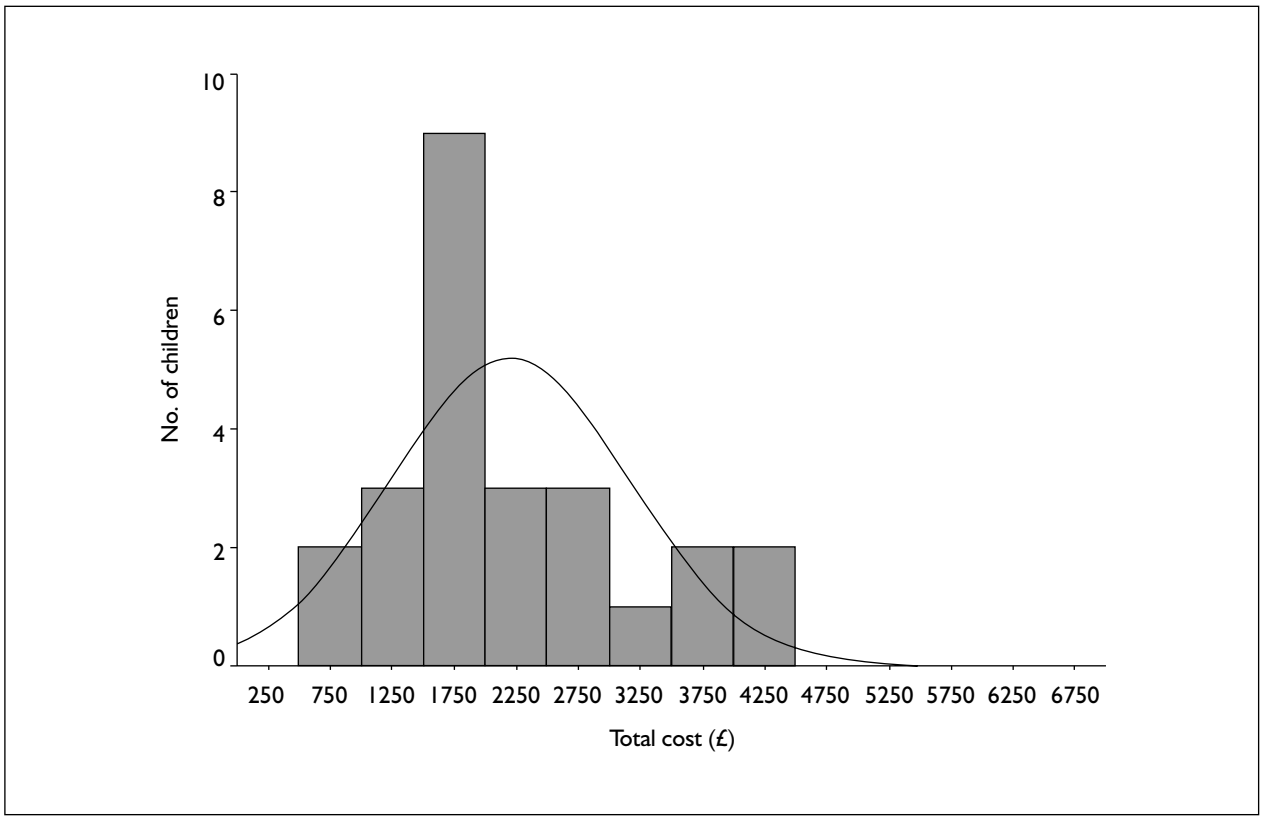

FIGURE 9 *Distribution and normal curve of total cost for each child in the PA arm of the trial*

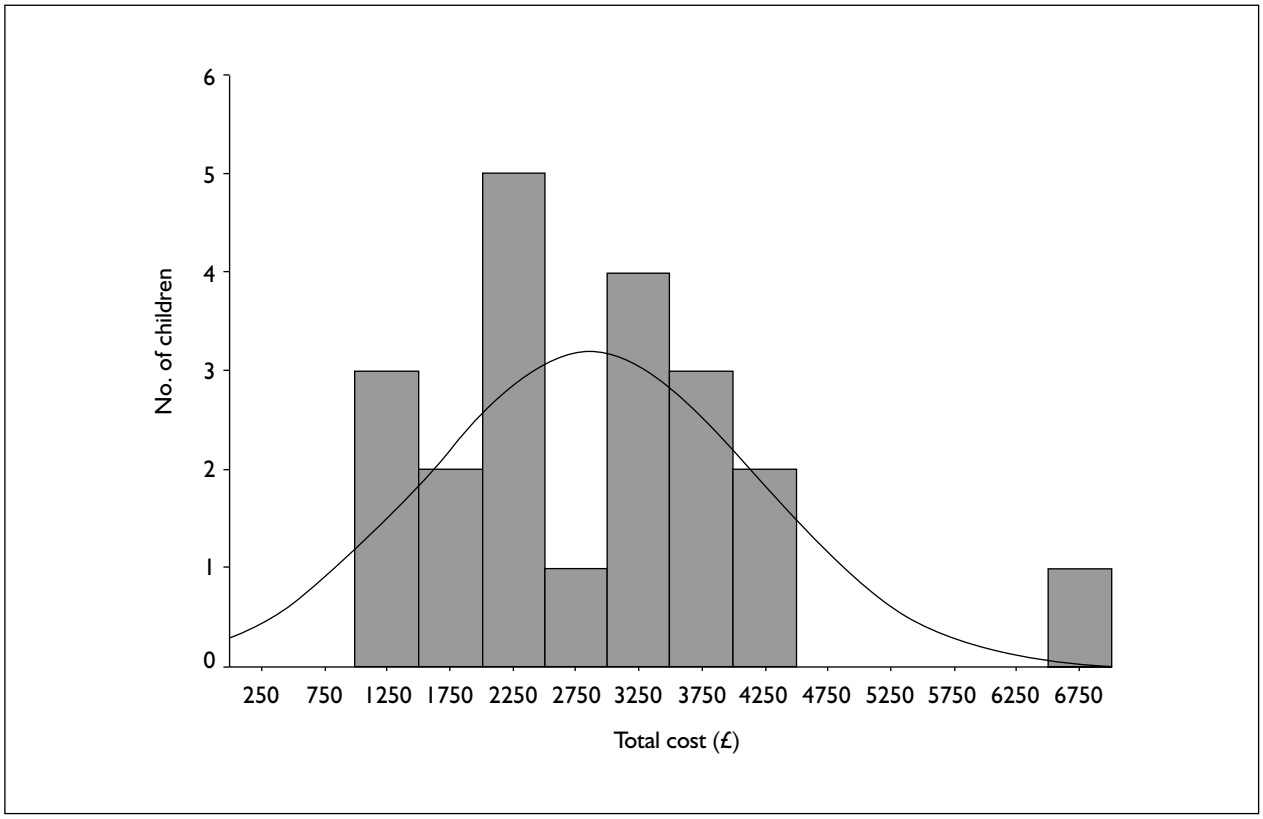

FIGURE 10 *Distribution and normal curve of total cost for each child in the FSW arm of the trial*

TABLE 17 *Mean (± SD) cost per child (£) during the 6 months intervention according to GMFM, MQ and CQ by quartiles*

Quartile	*N*	GMFM	MQ	CQ
1st (worst affected)	18	2783 (± 930)	2971 (± 1279)	2910 (± 1362)
2nd	19	2567 (± 1423)	2856 (± 1041)	2819 (± 1060)
3rd	18	2415 (± 1117)	2139 (± 1123)	169 (± 963)
4th (least affected)	19	1647 (± 1143)	1442 (± 837)	1960 (± 1095)

TABLE 18 *Mean (± SD) cost per child during the 6 months intervention according to CDC*

CDC	*N*	Mean cost (£)	SD (£)
Chester	10	1759	916
Liverpool	2	2377	657
Manchester	2	1210	1102
Ormskirk	6	1167	1350
Rhyl	2	1976	504
Warrington	8	1796	448
Whiston	2	4222	3389
Widnes	6	2121	918
Wigan	7	2384	986
Winsford	6	1884	1172
Wirral	20	3219	1114
Wrexham	3	2339	1083

The mean (±SD) cost of services for 15 children with diplegia was £2073 (±£1343), for 31 children with hemiplegia it was £2049 (±£975), and for 34 children with tetraplegia it was £2822 (±£1300). To consider further whether the most disabled children attracted the greatest expenditure, the GMFM, MQ and CQ scores were divided into quartiles. The results are set out in *Table 17*.

To see if there was any geographical variation, the cost was also calculated for each child according to the CDC at which treatment was based (*Table 18*). However, it should be noted that no account was taken in this calculation of the children's degree of disability.

Cost-effectiveness analysis

Cost-effectiveness analysis involved drawing up a balance sheet of the costs and consequences or benefits of a healthcare intervention, and comparing them with, in most cases, usual or existing patterns of service delivery.[69] It was important to note that a treatment or intervention could not in itself be said to be cost-effective. However, it could be said to be cost-effective compared with another mode of treatment or service delivery.

The perspective from which costs and consequences were measured and reported was also important. In this study of the cost-effectiveness of additional physiotherapy for children with CP, we took a predominantly NHS perspective. This covered staff costs (salaries, salary on-costs, training, overheads and capital overheads). Some costs which fell on the educational sector were included, as services received by children in the trial span a range of sectors. Travel costs were not included in this analysis.

Effectiveness: key clinical outcome measure

For the purposes of this cost-effectiveness analysis, the mean difference between GMFM at T1 and T2 was used. *Table 19* shows the mean effect and SD between baseline (T1) and post-intervention (T2) for the infants assessed for cost-effectiveness purposes in the three arms of the trial. For this analysis, an independent samples *t*-test was used to calculate whether the observed changes in GMFM score for each group were significantly different to each other. No statistical differences were found between the CG and the PAG ($p = 0.103$) or between the CG and the FSWG ($p = 0.749$).

Incremental cost-effectiveness analysis

An incremental cost-effectiveness ratio (ICER) allowed the calculation of the costs associated with switching from the existing treatment (the control treatment) to the intervention treatment. The ICER was obtained by dividing the cost differences (C2 – C1) by the effectiveness differences (E2 – E1) of two treatments, where C1 = mean cost per child in CG, C2 = mean cost per child in PAG, E1 = mean point score change on GMFM in CG and E2 = mean point score change on GMFM in PAG:

$$\begin{aligned}(C2 - C1)/(E2 - E1) &= (£2200.08 - £2087.90)/(14.48 - 9.47)\\ &= £112.18/5.01\\ &= £22.39\end{aligned}$$

The figure of £22.39 may be interpreted as the cost per child per unit of improvement on the GMFM scale of switching from the control treatment to the intervention treatment. The ICER indicates relative cost-effectiveness

TABLE 19 *Effectiveness figures based on the primary outcome measure, the GMFM score*[a]

Group	*N*	Mean GMFM at T1	Mean GMFM at T2	Difference ± SD[b]
Control	28	35.96	45.43	9.47 ± 11.32
PA	25	35.56	50.04	14.48 ± 10.56
FSW	21	36.62	45.10	8.48 ± 9.68

[a] Children were only included in this analysis if data were sufficient for a cost-effectiveness analysis.
[b] GMFM at T2 – GMFM at T1.

compared with any other existing treatment options.

Bootstrap point estimates and confidence intervals

As elsewhere in this study, bootstrapping was used. Here the statistical technique was used to examine uncertainty in the cost-effectiveness analysis.[70] Uncertainty may be caused by a small sample size or skewed cost data. In this trial, cost data were fairly normally distributed (see *Figures 8–10*), but sample sizes were relatively small.

Bootstrapping is a non-parametric method which simulates data by drawing participants at random from the database and then replacing them. This step was repeated until a 'pseudo data set' of 1000 trials was generated.[71] The pseudo data set was used to produce an estimate of the distribution of the cost-effectiveness point estimate. This was then used to derive the CI of the cost-effectiveness ratio.[71] Usual physiotherapy care (CG) was used as the base comparator service in the analysis.

PAG compared with CP

Figure 11 shows the cost-effectiveness plane, where each data point represents a cost-effectiveness ratio from the bootstrapping replication. Bootstrapping generated a mean ICER point estimate of £22, representing the mean ICER from the 1000 replications. Because in the raw data from the 74 participants no cost figures were less than £0, we calculated an upper one-sided CI instead of a two-sided CI, that is, we were only predicting the certainty of a given percentage of the bootstrapped population being below a certain upper limit. The bootstrap produced a 95% upper one-sided CI of £311.

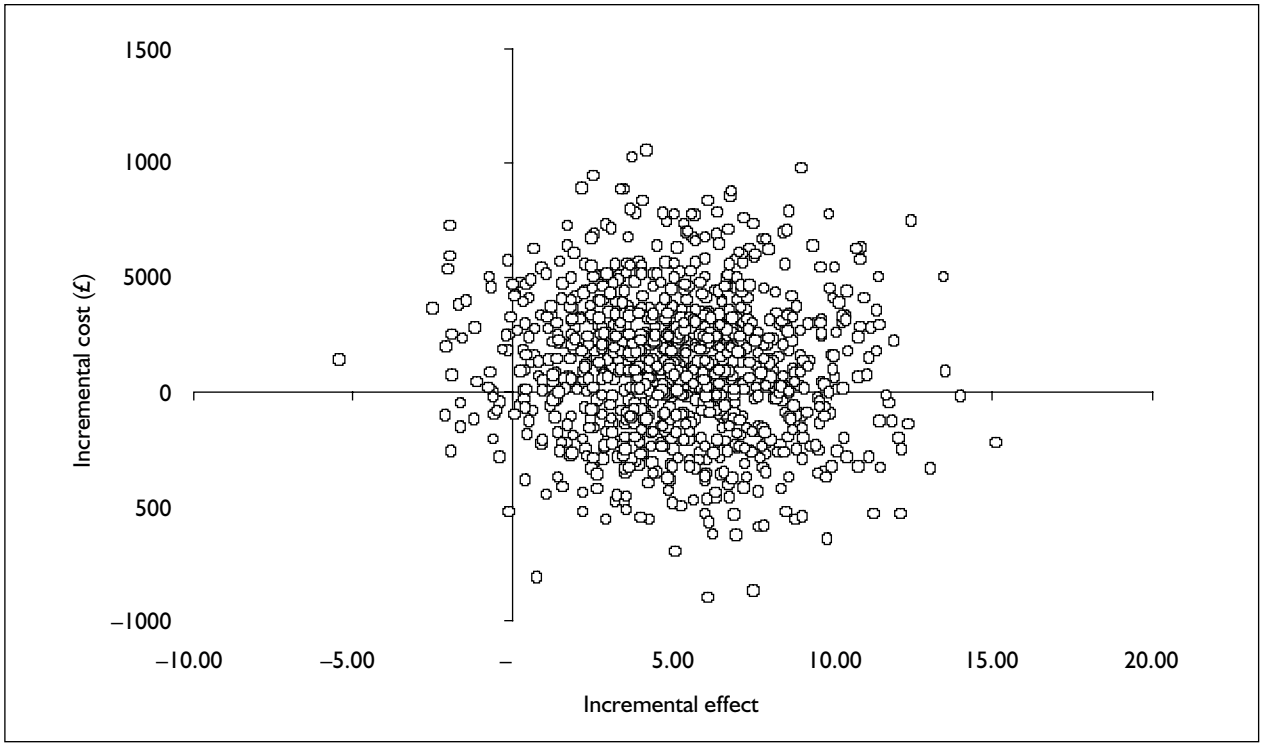

FIGURE 11 *Bootstrapped cost-effectiveness plane for PAG versus CG*

Interim discussion and conclusions

Comparing the PAG and the CG, the analysis showed that a one-point increase on the GMFM cost £22.39. A parallel economic analysis for the FSWG showed the FSW service to be more costly in terms of yielding benefits on the GMFM.

Overall, therefore, the benefit of a single point change was so small that it was not possible to conclude that additional physiotherapy by a PA for children with CP was a particularly cost-effective use of limited NHS resources when compared with usual levels of physiotherapy.

Multivariate analysis

Factors associated with child and family functioning

The aim of this analysis was to investigate which factors were associated with the services received by the children and with the predefined outcome measures, and whether the level of service provision had an impact on the outcome measures. Two sets of analyses were conducted. The first investigated relationships between the baseline measures and subsequent levels of service receipt during the intervention. The second sought to identify those variables, including levels of service provision, which demonstrated a relationship with change in outcome scores over the intervention period.

Services received

Two measures of the services received by the families were used (see Appendix 1 for further details). The first was 'Additional Services', indicating the range of services received by the family during the intervention period. This measure excluded the intervention and standard health services such as medical check-ups, health visitor visits and one-off assessment visits by a specialist or visits to doctors, but included such services a toy libraries and Portage in addition to therapies.

The second measure was the number of Non-intervention Physical Therapy sessions per week, also excluding the standard health visits. This was a measure of physical therapy received as part of services that were not part of the intervention. (The value for Non-intervention Physical Therapy was missing for one child, therefore an estimate was made from a regression on the number of physical therapy visits, the time elapsed between baseline and follow-up and experimental group membership). It should be noted that there was overlap between Additional Services and Non-intervention Physical Therapy because the Additional Services measure included services with a physical therapy component. (The two variables correlated at 0.69, indicating a large degree of overlap but some discreteness, i.e. different children and their families received different services.)

These measures related to services received during the intervention period. It was therefore possible that they were not independent of child and family outcomes during that period, that is, services may have been increased where outcomes continued to be poor or decreased where they were good. This was anticipated and we examined the ratings of contacts with services recorded by the independent assessor (see Appendix 1) at T1 baseline and T2 for the first 30 children. Over the course of the intervention there was a significant increase in services ($F1, 29 = 8.1$, $p = 0.008$) and this was similar for the three groups ($F2, 29 = 0.38$, $p = 0.687$). It appeared that being in one or other of the intervention groups had no effect. Inspection of levels of service failed to indicate any overall relationship with progress.

Statistical methods

The analyses were undertaken as a series of regressions in which each outcome variable at T2 was regressed against each predictor variable in turn, while simultaneously controlling for baseline scores on the outcome and the experimental design variables (group, area, nature of impairment, mother's education). Robust standard error estimators were used.

One purpose of the analysis was to look for previously unknown relationships. We therefore examined a wider range of potential predictor variables than was used for the RCT and longitudinal analyses. Tables report all relationships where $p \leqslant 0.1$, but to minimise the risk of Type 1 errors only results where $p \leqslant 0.01$ will be considered statistically significant for the purposes of interpretation.

Results

Relationships between service variables and baseline measures

Table 20 presents standardised β values and p-values for the relationships between the two service variables (Non-intervention Physical Therapy sessions and Additional Services) and the baseline values of all other variables. Only two factors associated at $p < 0.01$ with greater receipt

TABLE 20 *Summary of regression analysis[a] of relationships between service variables and predictor variables*

	Non-intervention physical therapy		Number of additional services	
	β	*p*	β	*p*
Design variables				
Area	0.63	<0.001	0.34	0.002
Diplegic impairment				
Tetraplegic impairment	0.26	0.01	0.22	0.03
Mother education to >16 years				
Mother education to >18 years				
PAG				
FSWG				
Other variables				
GMFM			–0.42	0.004
Vineland DL			–0.29	0.04
Griffith raw score	0.23	0.05	–0.27	0.08
PSI				
Family Needs				
Parent education				
Gender				
Family support				
COPE Adaptive				
Maladaptive coping				
HOME			–0.24	0.01
Family Cohesion	0.16	0.07		
Family Adaptability				
GHQ				
Internal LOC			–0.16	0.08
External LOC				
Life Stress				
Child Stress Index				
MQ				
CQ				

[a] Standardised β and *p*-values for each predictor, controlling for the design variables. Results are only reported where $p \geq 0.1$.

of Non-intervention Physical Therapy: area and tetraplegic impairment. The difference between areas was very substantial, with families from the Wirral receiving an average of 2.7 physical therapy sessions per week, compared with 1.1 for families elsewhere. Children with tetraplegia received on average 2.0 sessions, compared with 1.5 for children with diplegia and 0.9 for those with hemiplegia.

Factors associated with greater receipt of Additional Services at $p \leq 0.1$ were Area and lower baseline GMFM and HOME environment scores. Families from the Wirral area had a mean additional services score of 5.9 compared with 4.9 for other families. This suggested an average of around one extra weekly appointment with a healthcare professional.

Relationships between service receipt and outcomes

Regression was also used to investigate the impact of service receipt, plus other variables, on the way in which outcome scores changed across the intervention period. For this, intervention physiotherapy scores were added to non-intervention physiotherapy scores to make the new variable Total Physical Therapy.

As a first step in the analysis, the strength of the relationships between the child and family outcomes at T2 and their scores at baseline were examined. The correlations were 0.9 or above for all three child outcomes, indicating that the baseline scores for these outcomes very strongly predicted the scores at T2. For the Griffiths, GMFM and Vineland DL scores at T2, the

TABLE 21 *Summary of regression analysis[a] of relationships between outcomes and predictor variables*

	Child outcomes						Family outcomes			
	GMFM		Vineland DL		Griffiths		PSI		Family needs	
	β	*p*	β	*p*	β	*p*	β	*p*	β	*p*
Design variables										
Area									–0.22	0.003
Diplegic impairment	–0.08	0.05					0.16	0.02		
Tetraplegic impairment	–0.18	0.01	–0.13	0.01					0.15	0.08
Mother education to >16 years									–0.14	0.07
Mother education to >18 years							–0.18	0.01		
PAG	0.08	0.07								
FSWG										
Service variables										
Total Physical Therapy										
Additional Services	0.08	0.07			–0.06	0.06				
Other variables										
GMFM			0.17	0.10						
Vineland DL	–0.09	0.07								
Griffith raw score										
PSI	0.07	0.04								
Family Needs	0.01	0.10								
Gender										
Family support			–0.10	0.05						
COPE Adaptive			0.13	0.01						
Maladaptive coping										
HOME			0.17	<0.001						
Family Cohesion	–0.08	0.08					–0.16	0.09		
Family Adaptability					–0.05	0.07				
GHQ	0.11	0.002			0.06	0.08	0.23	0.01	0.16	0.05
Internal LOC									–0.16	0.02
External LOC			–0.12	0.04						
Life stress										
Child Stress Index										
MQ	0.30	<0.001			0.11	0.02				
CQ										

[a] Standardised β and *p*-values for each predictor, controlling for the design variables and outcome at baseline. Results are only reported where $p \leq 0.1$.

variations explained by the baseline score were 92, 86 and 80%, respectively. This left very little variation to be explained by other variables. Corresponding correlation coefficients for the two family outcomes were lower (0.8 and 0.67, respectively, explaining 65 and 45% of the variance, respectively). This suggested that these outcomes were more subject to change over time.

Table 21 shows that neither Total Physical Therapy nor Additional Services accounted for a significant proportion ($p \leq 0.01$) of any of the outcome measures after controlling for the experimental design variables and baseline outcome scores. A number of other factors, however, were significant.

Relationship between the outcome measures and other descriptive variables

Factors associated with GMFM at T2 were baseline motor functioning (MQ, $p < 0.001$), higher GHQ scores ($p = 0.002$) and (negatively) tetraplegia ($p = 0.01$).

Variables predictive of Vineland DL at T2 were better HOME ($p < 0.001$) and COPE scores ($p = 0.01$) and (negatively) tetraplegia ($p = 0.01$). No variables were significant at $p \leq 0.01$ with respect to Griffiths scores, although motor functioning (MQ) was close to significance at $p = 0.02$. Two factors were associated with parental stress scores (PSI): higher GHQ scores

BOX 1 *Case study: School for Parents (SfP) in the Wirral*

The 21 parents from the Wirral area with results at T2 all attended an SfP. This was set up by the local physiotherapists to support families of children with motor impairments. It was located at a CDC and provided physiotherapy and occupational therapy, hydrotherapy, toy library, parent-to-parent contact and support and, during school holidays, daily attendance, which provided much high levels of contact for parents who could visit the CDC. It was therefore amongst the most intensive and comprehensive support service for families of children with CP (based around physiotherapy services) in the region.

The mean birth weight of the children was 2.50 ± 1.00 kg (compared with a mean birth weight of 1.99 ± 1.01 kg for the rest of the children). Their gestation at birth was 36.3 ± 4.4 weeks (compared with 33.1 ± 6.8 weeks for the rest of the children). The male:female ratio of children who attended the SfP was similar to those who did not (1.4:1). Compared with the other 55 children who finished the intervention, they were diagnosed as having CP at a significantly younger age: 7.3 ± 4.0 months for the SfP children compared with 11.9 ± 6.8 months ($p = 0.001$). They were also younger at entry to the study: 14.7 ± 5.8 months compared with 21.3 ± 8.7 months. The parental social class was similar (6.6 ± 1.9 for those at the SfP compared with 6.6 ± 1.8 for the others). The mean MQ (± SD) was 58.3 ± 33.7 for the SfP children compared with 60.2 ± 26.7 for the others, and the mean CQ was 71.1 ± 33.3 for the SfP children compared with 76.0 ± 28.2 for the others. This indicated higher levels of motor and cognitive disability in the SfP group.

There were relatively fewer children with diplegia (10% of children in the SfP group and 25% in the remainder).

As a group, the 21 Wirral families who attended the SfP received significantly higher levels of total Physical Therapy sessions and Additional Services compared with families in other areas:

- Total Physical Therapy sessions: 56.5 ± 28.9 versus 23.6 ± 15.7 ($p = 0.000$)
- Additional Services mean rating 5.9 ±1.0 versus 4.8 ± 1.3 ($p = 0.002$)
- Mean sessions per week: 2.9 ±0.82 versus 1.3 ± 0.80 ($p = 0.000$).

In addition, eight (38%) of these 21 families also received Portage compared with seven (13%) of the remainder.

The families of children with CP who lived on the Wirral had a much higher input of services and had lower Family Needs at T2, but there were no other differences with the other families.

($p = 0.01$) and mothers who finished their education at 16 years ($p = 0.01$).

There was a strong association between geographical area and reduced Family Needs scores ($p = 0.003$): the mean Family Needs score for Wirral families decreased from 57.3 to 52.0 between T1 and T2, whereas the score for families from other areas increased from 55.6 to 56.9.

Details of a case study, the School for Parents (SfP) in the Wirral, are given in *Box 1*.

Interim discussion and conclusions

The analyses show that levels of receipt of physical therapy (in addition to that provided by the intervention) and of additional services were considerably higher in the Wirral than in other areas. In addition, the presence of tetraplegia significantly affected the amount of physical therapy received, whereas higher baseline GMFM and HOME scores were associated with lower levels of additional service receipt.

There was no evidence for a significant relationship between any of the five outcome measures and the two measures of services received. Interpretation of this result, however, was not straightforward: the service variables relate to services received between T1 and T2, and may not be independent of the outcomes themselves. A general trend for outcomes to modify service provision negatively would have reduced the likelihood of detecting any positive effect of services on outcomes. As previously noted, there was increased provision of services over the intervention period but there were no differences between the groups and no suggestion of a consistent relationship between outcome and the provision of services. Service provision was substantially higher on the Wirral, and the only association with outcome at T2 was Family Needs, which were significantly reduced for children whose care was based at the Wirral CDC.

A number of factors were associated with the child and family outcomes. For GMFM, these were baseline motor functioning (MQ), higher GHQ scores and (negatively) tetraplegia. For Vineland DL these were better HOME and COPE scores and (negatively) tetraplegia. No factors were significantly associated with Griffiths scores. Stress (PSI scores) was significantly reduced in families with higher GHQ scores and more highly educated mothers.

The qualitative data

This section describes (a) parents' satisfaction with the intervention and what they liked and disliked, (b) the nature of family needs in terms of the weekly diaries kept by the FSWs and (c) the question of whether there were subgroups of parents (cases) that had different needs or responded to the intervention differently than the rest.

Sources of data

There were five sources of qualitative data. These were the parent satisfaction interviews, the parents' diaries, the PA diary, the FSW diary and discussions with the PAs and FSWs. Further details of the measures are given in Appendix 1.

Parent satisfaction interview

Following the intervention period, the independent assessor asked all parents:

- what their overall impressions of the service were
- what they liked about it
- what they did not like about it
- what they would do differently or change about the intervention.

Their responses were written down verbatim or paraphrased. They were also asked to score their overall satisfaction with the intervention on a scale from 1 (very poor) to 10 (excellent).

Parent diary

Parents were asked to keep a daily diary over the intervention period about contacts with services related to their child having a disability. Contacts included healthcare services, provisions such as toy libraries, parent groups, Portage and nurseries. Sixty-six parents (22 in the CG, 21 in the PAG and 23 in the FSWG) provided reasonably complete diaries. Many not only recorded services received but also made comments about the service. These comments were used in relation to (c) above.

PA diary

The PAs recorded visits made or cancelled and what activities had taken place during the visit. The PAs' brief was to provide extra physiotherapy and so few made any comments about other aspects of working with the family in the diaries. These diaries therefore offered little help about the nature of family needs. However, from parent comments and discussions with the PAs, their role was more extensive for several families.

FSW diary

The FSWs recorded the nature of every home visit. The entries were far more detailed than those made by the PAs. This was appropriate as they had to define with the parents the actual help provided. They frequently documented who was present, what they were doing and the nature of the conversations that took place during this time, problems parents had, advice and solutions, feelings and other services received, as described and commented on by the parents. There was no preset format for the visits or the diaries and most were lengthy descriptions of the visits. These diaries provided the most informative qualitative data about the 'lives' of the families during this period.

PA/FSW discussions

During the study, regular meetings were held between the research staff and the PAs and FSWs. Often issues were discussed and the views expressed were recorded.

At the end of the study, they also expressed their views about what had been valued.

Analysis (also see Appendix 1)

The verbatim notes of the parent satisfaction interview and the diaries were analysed in the same way. Each was initially read through to provide familiarity with the nature of the text and an overview of the type of information that they contained. A note was made of any comments about satisfaction and additional services received. A number of themes emerged, particularly from the more detailed FSW diaries. Once these themes had been exhausted, each interview was re-read. Where the contents pertained to one of the themes, this was noted under the appropriate heading. This procedure was followed until the contents of all of the diaries were written up within the appropriate headings. Particular attention was given to examining the concordance between data sources. Comparison was also made between FSW diaries and the self-completed questionnaires of Family Needs, Family Support, Family Cohesion and stress, and in nearly every instance the two sources of information agreed. These analyses were carried out by an experienced researcher independent of the study.

Once the thematic framework had been established, a subset of the diaries and interviews was read by a member of the research team. Agreement was very high.

The contents within each separate heading were then read through and written up to provide an

overview of the findings. These were then amalgamated into a draft and quotations were included to add illustration to the text.

The summaries of the FSW diaries were also sent to the FSWs for comment on accuracy and whether any points/issues had been under- or overemphasised or missed. They agreed that the summaries below were accurate representations of their diaries.

Results

Parent satisfaction

Eight (29%) parents in the CG made the comment that they were disappointed at the group allocation because they had wanted to receive the additional physiotherapy. One mentioned that they had wanted to receive an FSW.

Forty (91%) of the parents in the intervention groups were satisfied with the intervention – the majority were very happy, whether in the PAG or FSWG. Many of the comments were effusive: "Absolutely brilliant", "Smashing", "Delighted". This was reflected in the rating scores. The mean rating of satisfaction was 8, with a range from 3 to 10. The mode was 10. Only two parents scored below the mid-point level of 5, suggesting greater dissatisfaction than satisfaction.

Four parents (two from the PAG and two from the FSWG) were negative about the home visiting. For the PAG, one mother and the PA failed to form a relationship, with comments such as "Didn't feel we had anything to talk about" or "no empathy". In the other instance, the parent felt that she had not received much support and was disappointed that eight sessions had been missed because of holidays and sickness. One mother in the FSW group stated that they did not miss the visiting once it had stopped, "as the child already does so much". This family had very high ratings for additional services.

There were several suggestions for improvement. One mother in the PA group suggested that the PA and the usual physiotherapist should work together more closely. Given that the intervention demanded close working between the PA and the child's physiotherapist, this comment was significant. However, it appeared to relate to one PA–CDC physiotherapist pair, rather than to be a general problem. Two suggestions emerged from several parents in the FSWG. First, some thought that the weekly visits were too frequent and that these should be made less often. Second, a number of parents thought that it would have been more helpful if they had had contact with an FSW from the initial diagnosis.

In response to the question, "what did you like about the home-visiting?", few in the PA group specified reasons, apart from the fact that they liked the PA herself: "Seen as a member of the family, they all love … coming", "the [PA] was very nice", "lovely". One parent stated that it was "Brilliant to have the extra stimulation", and another that the child's "Concentration and attention greatly improved". Two parents stated they appreciated the opportunity to talk to the PA:

> "Someone I could talk to – more help to me than actually doing things with [child]".
> "Helped not only [child] but helpful to discuss problems with [child], talking really helped".

This indicated that the PAs could not avoid providing 'emotional support' in some cases. This was also noted by the PAs themselves during the staff meetings and was estimated to be the case in around half the families.

Generally parents were "sorry it [the intervention] had to finish" and they missed the visits from the intervention worker.

Establishing an empathic relationship with someone they could talk to was also the most cited benefit by the FSWG. Parents felt that it helped that the FSW had a child with a disability herself as she had "had first-hand experience", "knew how we felt", "could understand our problems" and also "knew all the pitfalls". It was:

> "Marvellous to be able to talk to someone who understood how I felt. Before the family support worker, I had felt that no one really understood".
> "Good to have someone to talk to and share anxieties and moans".

Most parents liked and found it easy to talk to the FSW, e.g. "Lovely person", "XX is wonderful". Consequently, they "Missed the visits when they stopped" and for one mother, "I cried when she left – she was lovely".

Frequently noted benefits included providing practical information about benefits, helping parents to fill in forms, how to access activities and further support and help with dealing with other professionals. For example:

> "Helped us get extra benefit and put us in touch with Riding for the Disabled".

"Made sure we received all the benefits, put us in touch with support groups".
"Helped with the statementing process, better seating and mobility aids".

Some parents stayed in touch with the FSW after the project finished. In several cases, the families were perceived to be so vulnerable that the FSW and the families felt they had to keep in touch.

The FSW intervention

Twenty-six FSW diaries were available for analysis and included three from families who withdrew early from the intervention. All three felt they did not need any FSW support. In one family, the FSW diary recorded two visits during which the FSW played with the siblings and talked about the project. The family was already in touch with a number of services, including respite care, social services, the disability organisation SCOPE and Family Link, and the child attended special nursery every day. They informed the FSW that they had hoped for extra physiotherapy from the project and felt that they had no need for an FSW. The mother of the second family that withdrew appeared to feel that she "Knew everyone that she needed to know and if she didn't, she found a way of getting to know if it would benefit [child]". After the first visit, she rang to withdraw from the study. The third mother pulled out of the project after two visits because "She didn't get extra physio. Mum said she had enough people in her life for help". All three recorded they were well supported on the Family Support Scale and recorded low scores on the Family Needs Scale.

Mothers had by far the most contact with the FSW; even in cases where the father was available during the day, the appointment was made with the mother. However, there were three fathers who did talk about their feelings to the FSW (see p. 41) and another three who chatted generally with the FSW on one or more occasion.

Using all the qualitative information, the parents in the FSW group were categorised in the levels of help they needed:

- Three needed little or no additional help.
- Seven were happy to have some input but did not seem to have very great needs and stated the visits could be less frequent.
- Sixteen (62%) had more problems and consequently used the FSW to a greater extent. Ten of these mothers appeared to have the most need for the FSW and spent a lot of time talking over their feelings and problems: six discussed marital problems, two discussed family problems and two both sets of problems. A comparison of the Total Life Stress score for these 10 compared with the remainder of the group showed similar scores at T1 but considerably higher at T2, reflecting the increase in stressful life events during the 6-month intervention period.

Main themes

Two main themes emerged: practical and emotional support.

Practical support

A number of categories were clear in the practical support theme: practical help, providing information on services, giving advice and support on issues relating to the child's condition and acting as an advocate or go-between with other health professionals.

Practical help Although the FSW did provide practical help, this was not a dominant feature of their visits. Just 26 instances of practical help were documented in the diaries. These included bathing, dressing or playing with a child, or siblings, and were usually carried out while the mother got dressed, made a telephone call or cared for a different sibling, or to give the parents some valuable time together. There were one-off instances where the FSW did the ironing, took the child to the doctor and helped put a standing frame together. These were therefore occasional functions performed when something altered the mother's routine at the time of the visit.

Information role Many entries described the role of providing information about services. Indeed, at the first visit, the FSWs appeared to check that the family was receiving all their entitled benefits. The FSWs provided and/or helped to fill in claim forms and supply information on statutory and voluntary services. These included the Family Fund, Disability Living Allowance, Invalid Care Allowance (ICA), SCOPE, toy library, local schools, Educational Statementing and Social Services. One entry read:

"We spent the whole of the visit talking about [the] Mobility [allowance] and how it works. Mum didn't really understand it".

Another entry was,

"[Mother] crying in kitchen. Made her a drink. Her father had refused to take her to X, to pick [child] up. Sister still in hospital and can't drive for six weeks. Suggested she get in touch with social worker to ask if there's any help, or suggested she set up a contract

> with a local taxi firm so she won't have to rely on anybody. Also family fund that give out driving lessons".

The FSWs also handed out information leaflets from different sources, such as Hemi Help, and six parents were given information on the Statementing process for school, followed up with a discussion by the FSW.

General supportive role The FSWs were parents of children with CP. This came through in the diaries and they appeared to be a good source of information for the parents, sharing similar experiences. Parents usually discussed general issues about the child's progress each week: their general health, visits to health professionals, achievements, behaviour and so on. They also talked about more specific issues that affected them, such as using splints, sleeping, feeding, seating and adaptations, and management, for example: "[Child] lying on couch, suggested she try putting him in his chair for help with his breathing".

Parents also discussed particular anxieties such as ill health, lack of progress and the future for the child:

> "[Child] has been accepted into … school, which mother disappointed about. She has heard that this school is for severely disabled children. Mother and father visiting the school tomorrow. We talked it through and she now realises that at least [child] will be getting the attention and all the services that he needs and deserves".

Advocacy role There were many instances of the FSWs acting as a go-between with professionals. This role appeared to emerge in two ways. First, when the parent herself expressed a problem with a particular professional or service, and second, when the FSW identified a need to consult with a professional. Some parents expressed concern that a particular professional was not available or that there was a lack of input generally from the services. Problems occurred when parents either did not like a specific professional or were not happy with an aspect of care. These two areas were most frequently related to occupational therapy or the health visiting service, and occasionally to physiotherapy. The response of the FSW was usually to listen and chat. Sometimes practical ways forward were suggested, such as contacting the health professional to make another appointment, or discussing their concerns with the person concerned, and writing down what they wanted to ask the person beforehand. Other suggestions included requesting a different health professional to care for the child. There were three instances where the parent requested that the FSW contacted the professional on their behalf. These were talking to a physiotherapist about obtaining shoes to fit over the child's splints, speaking to a social worker to discuss ways in which she could help the family and being present when the health visitor came. One FSW also attended a case conference with the mother (at the mother's request) that was aimed at putting a care plan in place.

There were instances when the FSW had concerns about the child or family and suggested consulting with a professional. This included advice to contact the GP, in one case when a child had lost the use of her arm and leg for a few minutes, and another when the child was not looking well. One mother was encouraged to contact the physiotherapist immediately rather than wait for a visit when her child kept falling over. Similarly, another child was having many fits so the FSW suggested contacting the epilepsy nurse. On another occasion, the FSW was concerned that the child had a hearing impairment and asked the mother if she had noticed anything, suggesting that it would be worth mentioning to the consultant. Three mothers repeatedly missed appointments, either for themselves or their child, and the FSW tried to explain the importance of these appointments and to encourage them to rearrange the appointment.

Emotional support

Seventeen (65%) of mothers chose to disclose very personal information relating to matters such as their feelings of depression or marital problems. Often these recurring themes were discussed over several visits. The provision of emotional support, particularly to the mothers, appeared to be a major help. It was striking that some of the mothers disclosed very personal information early on in the visits, sometimes even in the first one: "I was surprised how much she disclosed on my first visit, and I felt she really wanted to talk and was very open". This suggested that some families/mothers had a great need to talk about their problems and that the FSW provided the means by which to do so. The following two entries illustrate the extent to which some mothers felt supported by the FSW:

> "Mother asked by health visitor to go once a week but mother said FSW visits were more beneficial and she can talk to me".
> "Mother had a big surprise for me – has come off the Prozac – said just talking to someone had lightened her load and she thought she'd have a go without it".

The main topics/problems related to the relationship with the partner, family issues, the mother's dissatisfaction with her life, financial concerns and major life events. This group reported more life event stressors than the others on the standardised questionnaire.

Problems with partner Eleven mothers talked over problems about their relationship with their partner. Indeed, during the course of the 6-month period, four couples told the FSW that they had split up. This was reflected in a higher score on the life stressor scale under the category of separation. On this scale, six couples in the FSWG reported a separation compared with two amongst the rest of the participants.

One mother spoke of her marital difficulties on the first FSW visit and others also disclosed relationship problems early on in the 6-month period. These included a mother having unprotected sex with the partner from whom she had already split up and a father being annoyed with the mother because she could not have sex following an operation. Two mothers told of their feelings about their partner going out on his own or to the pub every night, one of whom was lying about where he was going. Another mother spoke of how she wanted to work but her partner wanted to have another child. A different mother felt that she had little time for herself yet the father had taken a week off work to play golf. Another entry read, "Mum worried about dad, he doesn't talk about B … having CP, mum worries that he is bottling it all up. She is worried that with the strain of their problems they may fall apart".

Seven (27%) of the 26 mothers also spoke of their feelings of depression or inability to cope. One diary entry read, "Mother on her own today, really opened up – feelings of emptiness since losing the baby, wants to try for another". Another in the same diary read, "Mother has had feelings of desperation where she just wants to end it all. I suggested seeing a counsellor – she said she talks to me".

An entry in a different diary read, "Today was a deep day, very private issues were brought up and discussed".

There were no further details of this in the diary. But later in the same diary: "Said she wanted a divorce – this led to a major disclosure!" Again, the nature of this was not disclosed. Other issues for this particular mother were her feelings of "lack of control", "deep depression", "wanting to feel of being a person in her own right and not just a mother". She also felt, "The child had driven a wedge between all the family. Friends don't come round any more". In response, the FSW listened and explored how the mother felt about this. She also gave her a book on siblings, which she thought might help. During the 6-month period this mother and her partner split up but got back together shortly after as they were "giving it a go".

Three fathers opened up somewhat to the FSW. One diary entry read, "Father spoke at length regarding his problems, no job, partner could be pregnant, got the feeling he needs to talk". A later entry read, "Father subject to child abuse when younger, explored this and his feelings". Another father spoke about his feelings since the death of his child and worried that he could not afford the funeral.

In addition to problems with the partner, parents also spoke of difficulties with their extended families. This sometimes related to a lack of support in understanding the child and his/her condition. For example:

> "[Name] believes husband's mother does not really accept J's disability, she thinks he's going to get better and that eventually he will be normal".
> "[Older sister] keeps comparing her child who is a similar age to H. This upsets mother".
> Another diary read, "Mum distressed that aunt always introduces C as 'the child with CP'".

Life events During the course of the 6-month period, some families experienced major life events which they chose to talk over with their FSW. These included the death of their child, a miscarriage, adoption, major operations, an abortion, splitting with a partner, moving house and being in financial debt. This was reflected in a higher score on the PSI Life Stressors variable, compared with the other two study groups.

In the tragic instance of the death of the child, both the mother and the father turned to the FSW for emotional support. They asked her to continue the visits and invited her to the service. The diary entry for the day of the funeral included the following: "[The parents] had given the vicar a list of people to thank, I was included as a special family friend".

In one instance of marital breakdown, the mother turned to the FSW for both emotional and practical support. As well as listening to the mother's outpourings, together they "came up

with a sort of a plan" for how she would cope on a day-to-day basis. They also discussed options for living arrangements and contacting a social worker regarding financial aid.

Valued support The following instances illustrate the extent to which the FSW became valued by the families. Two entries told of the mother, or mother and father, buying cakes for the FSW to share on their birthday, while another mother bought the FSW a birthday present. Many spoke of how they valued their FSW's support: "Was waiting for me to come all week", "Loves me coming". When it came to the end of the project, one mother was "Crying when I left". One father had "Taken the day off to say goodbye and to thank me for all that I have done for his family". Altogether three families asked if the visits could be extended (this included one who requested this just 3 months into the study period), and seven mothers asked if they could keep in touch or remain friends.

The independent assessor also made some notes during the parent satisfaction interview, giving testimony to the help many families had received, and in all instances these corresponded to the FSW diaries. Comments on some of the PAG diaries suggested that families in the group had had similar problems.

Interim discussion and conclusions

There was strong concordance between the different sources of data. Most parents found the intervention, whether by PA or FSW, to be beneficial. One of the most striking findings was the apparent need to talk and to have someone listen to emotional problems. This was evident from reading the FSWs' diaries. It probably also existed in the other two groups since there was little difference between groups as measured by the family, stress and coping measures. During the debriefing meetings the PAs described having to talk with mothers. Also, during the parent satisfaction interview, the independent assessor noted that there were two parents who stated they valued the nature of their relationship with the PA rather than the child's therapy.

For many of the parents, problems with their relationships or daily routine were associated with problems caused by the child's disability. However, for many others, coping with the child's condition itself was not the focal point: there were other problems related to lack of support, marital difficulties and disputes with family members.

Comparison of families in the FSW group with the other groups did not reveal any major differences in the descriptive and measured variables, apart from increased Life Stressors. Hence it could be reasonably assumed that this level of need also existed in the other groups.

Three of these families are described in more detail in Appendix 3 as case studies to illustrate the type of issues they faced and the nature of the help that they required.

Chapter 4
Discussion

The study aimed to (1) investigate the effectiveness of increasing the intensity of physiotherapy on motor functioning in preschool children with spastic CP, (2) explore the role of the physiotherapist working with young children with CP, and (3) provide broader understanding of the families of children with CP. The underlying reason for the first aim was a commonly held belief that the more physical therapy given to children with CP, the better their future development and quality of life would be. This belief led to the study's second aim, which was to explore the physiotherapist's role. It addressed the research question of whether the physiotherapist's primary focus was to provide physical therapy or whether, as claimed by some physiotherapists, it should include a broader view of the needs of the child and take into account family functioning. As described in Chapter 1, early intervention programmes for children with disability have generally moved towards a model that is family focused and away from one which is focused on therapy and limited to the child.

This investigation was designed as a multicentre RCT with a 6-month intervention period and follow-up at 12 and 18 months. The study design separated the effect of extra physiotherapy from family support. Randomisation was to three groups. One group received extra physical therapy from a PA to test the efficacy of increased physical therapy alone. A second group received support from an FSW to explore the effectiveness of focusing on family needs. A third group, receiving standard services, acted as a CG to evaluate the effectiveness of the interventions. Each intervention was applied for 1 hour each week at home over a 6-month period.

A wide range of variables was measured to provide a broad understanding of the needs of families of children with CP. These were prespecified and included the collection of information about the nature and levels of services received by the families and their cost implications.

The first part of this chapter is about the efficacy of increased physical therapy. The second part is about the efficacy of the family support worker. The third part focuses on the nature and needs of the families, services provided and implications for the role of the physiotherapist.

The effect of additional physical therapy by a physiotherapy assistant

This was tested by assessment at the end of the intervention (T2) and at the 6- and 12-month follow-ups (T3 and T4). There was no evidence to support:

- *Hypothesis 1*. Extra home-based physical therapy by a PA improves motor function in the child with spastic CP.
- *Hypothesis 2*. Extra home-based physical therapy by a PA improves general development of the child with spastic CP.

General child development (Hypothesis 2) was measured by the Griffiths and Vineland scales and these unequivocally showed no difference as a result of the intervention.

The primary measure of motor function (Hypothesis 1) used in this study was the GMFM. There was a mean 5-point advantage in the GMFM score for the physiotherapy assistant group (*Table 9*, p. 21). The unadjusted analysis was not significant ($p = 0.09$), but marginal statistical significance ($p = 0.04$) was found on the adjusted analysis by the bootstrap method [see the section 'Gross Motor Function Measure (primary outcome)' (p. 21)]. Most importantly, this average effect size was not only considerably less than the 14 points set as the original criterion for the size of change that would be considered to be clinically important, but the upper 95% CI was also 4 points less.

Since there was some statistical uncertainty in relation to the GMFM result and the final sample size was less than specified by the prestudy sample size calculation, there was concern that a Type 2 error might have occurred. There are, however, several reasons for thinking that it is reasonable to be confident that the physical therapy intervention had no real effect. First, the observed mean change of around 5 points had an upper 95% CI of 10.4,

which was still considerably lower than the preset target value of 14 points. Second, a *post hoc* power analysis using data from the study indicated that the sample size that was achieved gave the trial 80% power to detect an 8-point change in GMFM and 99% power to detect the clinically important change of 14 points, if such had existed. Third, despite randomisation, the PAG were slightly younger and had higher mean MQs and CQs than children in the other two groups. These differences were not significant, but raise the possibility that the children in the PAG were developing faster than those in the other groups and would therefore have been expected to continue to develop faster during the intervention period. Both covariates used in the main analysis (the GMFM score and the pattern of spasticity) defined the children's state at the point of entry to the study but not their rate of motor development. This was measured by the MQ. When this variable was used as an additional covariate in *post hoc* analyses, the mean effect size reduced to around 3 points (95% CI –1.5. to 7.9, $p > 0.09$), supporting the notion that the children in the PAG were less motorically disabled. Fourth, the multivariate analysis showed that the baseline scores at T1 were the main predictors of the child's outcome and accounted for 92% of the variance. In other words, the child's condition and level of development at the start of the study were the main factors that influenced outcomes after the 6-month intervention.

The conclusion that the additional physical therapy had little meaningful effect is also supported by studies published since the start of the present study. Bower and colleagues[23] used the GMFM as an outcome measure with a convenience sample of 56 children with bilateral CP, whose ages ranged between 3 and 12 years, to investigate the effects of intensive NDT therapy in a 6-month period. They found just a trend towards a significant improvement but any advantage declined over the next 6 months during which therapy reverted to its usual amount. Butler and Darrah[72] carried out a systematic review and concluded that NDT was not associated with better outcomes in most studies and that more intensive therapy produced no greater benefit. One exception to these reports of no clinically or statistically significant effects was a study by Tsorlakis and colleagues[73] with proportional stratification for age, sex and type of CP. Thirty-four children aged 3–14 years were assigned to two groups. Seventeen children had NDT twice per week and the other 17 had NDT five times per week. Using a re-standardised version of the GMFM, the change scores for children in the intensive group were significantly higher than those for children receiving less intense therapy. However, their analysis can be criticised for the use of *t*-tests rather than ANCOVA, with no correction for multiple tests. Finally, a recent systematic review of the effects of early intervention on motor development of infants considered to be at high risk for, or with, developmental motor disorders analysed 12 high-quality studies of NDT and also found no beneficial effect on motor development.[74]

Therefore, at this time, most studies support our conclusion of little meaningful effect for intensive physical therapy intervention in terms of attainments on motor functioning tests and increased levels of development.

However, the maintenance of the GMFM score must also be seen as a positive outcome for children who are severely affected by spasticity and whose motor function might be expected to deteriorate with time because of the development of fixed deformities. Inspection of the current data showed that only three children had later scores that were lower than the baseline scores. This might have indicated some deterioration, although a change in score could have also been affected by other factors on the day of assessment, such as ill health or less cooperation. Studies are required that investigate efficacy of treatment using motor measures that target individual needs precisely and include prevention of deterioration.

The effect of support from a family support worker

- *Hypothesis 3*. Extra home-based intervention by an FSW improves functioning of the family of a child with spastic CP.

Family functioning was measured by the maternal stress scores on the PSI and Family Needs scale. There was no change in these two primary outcome measures at T2 after 6 months of the intervention for the families in the FSWG and no difference between groups. Furthermore, the scores from the FACES, which measures several aspects of family functioning, showed no significant change over the 6-month period for children in all three groups.

The results of the follow-up analyses at T3 and T4 should be treated cautiously because of reduced numbers and those remaining in the study tended to have lower Family Needs scores and higher

HOME scores at T1 (*Table 11*) (see p. 23). This was similar for all the groups. There were no differences for the PSI at T3 and T4 or for Family Needs at T3. However, at T4, Family Needs were significantly reduced by 12% in the seven remaining families who had received FSW support (*Table 11*). However, this result in a considerably reduced group cannot be considered reliable. Therefore, we concluded that there was no evidence to support the hypothesis. This apparent lack of effect is similar to previous studies reviewed in Chapter 1.[38,40] However, the qualitative analyses showed that many parents felt they had benefited from the interventions.

The qualitative data and consideration of the quantitative measures used

Although the quantitative analyses showed no effect by either the FSW or PA, the qualitative data and the parent satisfaction measure indicated that most families were positive about the intervention (whether by PA or by FSW) and felt they had derived some benefit. For many, even those in the PAG, the main effect was perceived to be through the establishment of a trusting relationship with someone and feeling supported. The next most cited benefit was provision of information such as getting advice about access to benefits and medical treatment.

One reason why parental satisfaction was the only outcome measure indicating some effect may have been a lack of sensitivity of the other outcome measures. This appeared to be unlikely, for the following reasons. Motor ability and general development were assessed by three separate outcome measures. All were standardised and had been used in similar studies. The main measure, the GMFM, is a criterion-referenced rather than normative test, developed by physiotherapists in order to measure small changes in children with CP. By contrast, the Griffiths and Vineland tests are both normative tests devised to assess the development of normal populations, but we used the raw scores as outcome measures in order to increase sensitivity to change. No significant differences were found between the groups on these normative tests after the interventions, and the small difference found on the GMFM disappeared when subjected to more sophisticated statistical analysis.

Similar considerations may be applied to the family measures. The PSI is a test that had good validity and reliability. In the present study, it provided a wide range of scores, suggesting adequate discrimination between families with high and low levels of stress, and it correlated with expected variables of family cohesion and adaptability. The Family Needs Scale was not a psychometric measure with standardised scores, but it covered the range of needs, discriminated between families, had high face validity and correlated as expected with measures such as the PSI (parent and child domains), GHQ, Stressful Life Events and Family Cohesion.

Hence these outcome measures should have picked up any significant signs of improved functioning. The qualitative analysis, especially of the FSW diaries, strongly indicated that some families derived benefits concerned with emotional and inter-relational problems, and information needs. However, such benefits were not reflected in the group scores on the formal family measures and we were unable to demonstrate any benefit for the child. This may be because the most perceived benefit was by a small group of very vulnerable families whose stress levels and needs were high and remained high. Studies with more precise targeting of family needs and goal attainments are needed to explore the effects of specific family interventions.

Relationships between child and family measures

The salient finding was that none of the measures of the severity of the child's impairments (GMFM, Vineland DL and Griffiths scores) was significantly ($p < 0.01$) associated with family outcomes [parental stress (PSI) or Family Needs].

The Child Stress Index, which indicated the perceived stress that parents feel the child to be at the source of, was not correlated with any measures of severity of the child's impairment. Furthermore, no significant associations were found with the key variable of family cohesion. These data indicated that focusing solely on the child's level of motor functioning by providing physical therapies was unlikely to make any impact on families needs and levels of stress. Hence a more family-focused approach is required.

As expected, high parental stress (PSI) was associated with high scoring on the GHQ, which was designed to detect mental health problems including anxiety and stress, and a lower maternal level of education. However, a high score on the GHQ was also strongly associated with a higher level of physical functioning (GMFM). This latter

result was difficult to explain as it contrasted with other results. It was possible that there was a subgroup of parents whose children had significant motor disabilities but mild cognitive disabilities, causing increased stress in parents. Again, the implication would be for family-focused assessment to arrive at the complex individuality of children and families.

The qualitative data from the FSW diaries confirmed the view that some of these families benefited from the intervention: they felt that they had been supported and that some of their problems had been solved, although they were still left with high levels of unmet needs. Sloper and Turner recorded similar findings in their study of the families of children with severe physical impairment.[33]

Hence, as found in much of the literature on families of children with disabilities (e.g. Meisels and Shonkoff [75]), the severity of the child's condition was not a direct source of parental stress or family need. In this study, there was no evidence of a strong link between the severity of the child's impairment and family outcomes. Any stress resulting from the child was mediated by family resources. Yet, as discussed in the next section, the allocation of services appeared to be child focused rather than family focused, and raises questions about the role and training of the paediatricians and paediatric physiotherapists.

Levels of service and outcome variables

The study was set against a background of considerable support from various services. The analysis of services showed that these varied considerably between children, and also in some areas, such as the Wirral, there were very high levels indeed.

This raised two research questions:

- *Research Question (1)*. Are there identifiable factors that determine the distribution of services for families of children with CP?
- *Research Question (2)*. Does receiving these services make any difference?

Research question (1). Are there identifiable factors that determine the distribution of such services?

The two main measures, Non-intervention Physical Therapy and Additional Services (described in detail in Appendix 1), were similar for all three groups. It is particularly relevant to note that there was no difference between the PAG and FSWG (*Table 20*). Over the course of the study, the level of services increased equally for all three groups, probably reflecting the increasing access to services during the preschool years as children get older. Because the same data sources were used, there was overlap between Additional Services and Non-intervention Physical Therapy, which correlated at 0.69. However, there was also some discreteness, that is, different children and their families received different services (*Table 14*). Multivariate analysis showed that the children most likely to receive non-intervention physical therapy were those with tetraplegia (and those who lived in the Wirral), but there was no association with other measures of severity of the child's impairment. This was also reflected by the cost-effectiveness analysis, which found that children with tetraplegia and those most disabled attracted the highest cost (*Table 17*), which also showed a variation in cost with area (*Table 18*).

Looking at the Additional Services measure, the child variables (GMFM, Vineland and Griffths scores) suggest that children functioning at lower levels attracted additional services, although the level of disability (as indicated by MQ and CQ) was not associated with additional levels of services received. Less disabled children might have been more likely to access a wider range of facilities than those who were more disabled; our measures were not sufficiently sensitive to investigate this. Future studies need to look more carefully at the relationship between individual child's needs and services.

The other predictor of level of additional services was the HOME score. However, the distribution of the HOME data was skewed. Seven families had low scores and the children of these families had lower GMFM, MQ and CQ scores, although none were significantly different from the rest of the group. Since HOME assessed the child's environment (play opportunities, toys, responsiveness of mother), it was possible that at least some of these families had low scores because of the level of disability of the child and therefore that it was the low functioning level of the child that determined the score, rather than the environment.

There was wide variation in the allocation of services, as might be expected with a heterogeneous sample of children with CP. The variation in services was reflected by the spread in

the amount of services received by each child (*Table 14*) and the consequent costs for the 24-week period, which ranged between £250 and £5750 for children in the CG. For example, the strongest predictor of services was the area in which the child lived, exemplified by the children in the Wirral who attended the SfP, which provided intensive services.

It was of interest that there was no demonstrable association between measures of severity of the child's condition (MQ and CQ) and any of the family measures with the level of services provided. This may be a reflection of a lack of sensitivity of the measures of service input. Alternatively, it may be that what is lacking is a structured analysis of the needs of the child and family and therefore poor concordance between individuals and the provision of appropriate services, as suggested by the qualitative analysis. This is an area for further research.

Research question (2). Does receiving these services make any difference?

In this analysis, physical therapy services included all intervention visits by a PA to give a total figure for physical therapy. Children with CP generally received a high level of service input. Qualitative analysis of the diaries revealed that there were some parents who would not have minded if visits had been less frequent. Furthermore, the multivariate analysis (*Table 21*) showed no significant relationship between the two measures of service intensity and either child or family outcome measures.

The question was also examined using the case study (Box 1, p. 36) and the multivariate analysis (*Table 21*). Families who attended the SfP in the Wirral were compared with the rest. When their outcome was measured at T2, there were no differences for the GMFM, Vineland DL, Griffiths score and parental stress. There was a strong association ($p = 0.003$) between geographical area and a reduced Family Needs score: the mean Family Needs score for Wirral families decreased from 57.3 to 52.0 between T1 and T2, whereas the score for families from other areas increased from 55.6 to 56.9. However, this reduction in family needs may be of little clinical importance as it represented a reduction of only 1–2 needs compared with a mean of about 19.

In addressing the question of the efficacy of intensive physical therapy for the child with CP, one must first ask whether the extra hour of intervention each week could be expected to make a difference against this background of high input. This question cannot be resolved using the present data. Second, there was no relationship between the levels of physical therapy input and child outcome measures. This seriously brings into question the notion that increased intensity of intervention is effective and suggests that a considerable amount of the physical therapy provided may be unnecessary. Put another way, it seems likely that there is a sufficiency of therapy that will be helpful and above that there is no effect.

Methodological difficulties for this type of study

The main limitations of this study (reduced sample size and attrition of the sample for the longitudinal analysis) have been discussed previously. Like other studies of interventions for children with CP, it was difficult to achieve the number of participants initially projected. The reasons for this are recorded in Chapter 2 and the actions taken to attempt to remedy this are recorded in Appendix 4. There are many possible reasons why recruitment to such research studies is increasingly difficult and this would form a separate area for enquiry.

A second issue is how to cope with such a relatively rare condition and heterogeneous population. The prestudy estimate of the prevalence of CP included all forms of the condition. The study itself, however, attempted to achieve some homogeneity in the study population and was confined to children with spastic CP of perinatal origin. Hence the incidence of the condition studied may have been overestimated.

Another issue relating to the heterogeneity of this population is the possibility that some of the less severely affected children were not referred for inclusion in this study. Nevertheless, other descriptive variables and the levels of family stress in the present sample were as expected from the available literature. The reviewers therefore believe that this sample was reasonably representative of the families of young children with spastic CP of perinatal origin and their families in the UK.

Finally, the issue of whether the RCT is the most appropriate design for such an investigation may be questioned. The RCT is considered the gold standard for assessing clinical interventions.[70] However, this style of evaluating a therapy was

developed for drug studies. In such cases, the intervention is uniform – the nature of the drug and its dose are preset – the condition to be treated is fairly uniform and the sample size is large enough to offset heterogeneities. The present study draws attention to the fact that the RCT may not be an appropriate approach when an intervention is focused on a heterogeneous population such as young children with spastic CP of perinatal origin. Despite randomisation, children in the PAG had slightly higher MQs and CQs at T1, and children in the FSWG had slightly more visits to health services (suggesting more ill health) and their families had higher Family Needs scores. Moreover, a variety of factors have been shown to influence the development of young children and how families deal with disability and engage with the child or with help. In addition, the families and their children may also be receiving support and interventions from other sources in addition to those specified by the intervention study. These variables cannot be controlled for, although one might expect that they would be equally distributed by randomisation in a sufficiently large study. Siebes and colleagues, who examined 50 studies between 1990 and 2000, also concluded that single case studies, combined with efforts to develop measures specifically for children with CP and with high sensitivity, might make more valuable contributions to the scientific basis of therapeutic interventions for children with CP.[76] Butler and Darrah[72] reported a systematic review of the effects of NDT for children with CP and highlighted many of the same design criticisms as in Chapter 1 of this report, i.e. small numbers of participants with considerable heterogeneity, lack of control or comparison groups and non-randomised designs. They emphasised that CP is a fairly rare heterogeneous condition and that NDT is not a specific treatment delivered in a standardised manner. Furthermore, a child's family cannot be standardised. All these factors make controlled designs difficult. Blauw-Hospers and Hadders-Algra,[74] in a recent systematic review of the effects of early intervention on motor development of infants, concluded that it was likely that different interventions were appropriate for children of different ages. With these considerations in mind, the present study used three other methodologies in addition to the basic RCT. Apart from using additional validated measures of family functioning to feed into a multivariate analysis of the five main outcome measures, a case study approach was used to investigate the effect of more intensive input, and a qualitative analysis of parents' views was undertaken. Together with the information about services supplied to these families, these different approaches provided a rich supply of data about the way in which children with CP are treated in the UK. The experience of this present study also led to a conclusion that an RCT might be inappropriate for the assessment of interventions where the condition is complex and the setting is variable. In this case, both the family setting and the variable services received by different children contributed to the complexity. This point was also made in a report by the Medical Research Council on health technology in surgery in 1992[77] and a recent HTA report.[78]

Conclusions

No support was found for the proposition that increased intervention by a PA had any clinically important effect on the development of young children with CP. This may have been due to the high level of physical therapy support that children already received from local services. However, examination of the relationship between the intensity of services provided and the child's physical and general development also failed to show any benefits on the child outcome measures, as did a case study comparison of those receiving the highest level of services. These observations suggested that there may be a level of therapeutic input that is necessary and sufficient to maintain optimal progress, and that further effort beyond this is not justified. Biological factors probably set limits to what can be achieved. This requires further research.

The costs of general services for each child ranged from a few hundred pounds to over £6000 pounds for the 6-month period investigated. These costs were highest for the most disabled children, an observation which suggested appropriate allocation of services. However, concordance between the services received and the child's and family's needs was not obvious. There was a clear relationship between the pattern of CP and the provision of services, in that children with tetraplegia received most. However, this relationship disappeared when other measures of physical function, level of disability and family needs were used.

One area with a very comprehensive support system – the SfP – was the most expensive in terms of provision per child. There was, however, no objective evidence that the outcome for children and families receiving this very high level of

services was significantly better except for a slight reduction in family needs compared with the other areas.

The implications are as follows:

1. Research is needed to examine the issue of what the 'sufficient' levels of provision are for which children. These are likely to come from a variety of sources both from within and outside the NHS.
2. Research is needed to examine more directly the range and nature of support for these families. There should be specific focus on duplication and the factors or criteria that are used to decide on service provision for individual children and families.
3. From the family support perspective, there were only small group effects on family needs and reductions in parental stress. However, the qualitative analysis indicated large effects for some families, mainly those with high stress levels and additional medical, social or relational problems. These results were very similar to those reported in the literature on early intervention and family functioning. The general conclusion was that such services have to be tailored to individual family needs and situations.

Children diagnosed as having CP are referred to a CDC for physical therapy services, but the review results suggested that these services were largely child focused rather than being family focused. The reviewers did not find any significant provision of family support, other than the SfP, which appeared to facilitate contact between services and referral to other services. Those families which appeared to benefit from the FSW (as evidenced by the qualitative studies) clearly displayed great need, but received little input from other sources. However, as noted in Chapter 1, many physiotherapists believe that they have a dual function, offering support to parents in addition to physical therapy for the child. The present evidence suggests that support to families was relatively superficial. In this study, there was no evidence of referral to social services or for psychological support for many families identified as being in need of such referrals.

The overall criteria for the allocation of services to children and their families were not clear. There is, however, a natural tendency amongst parents to search out as much treatment as is available and a natural desire by clinicians to meet this demand. This was described by Paine in 1962[18] and 40 years later by Parkes and colleagues.[79] It is not unreasonable for parents to believe that more is better as far as physical therapy services are concerned, and this perception may be picked up by clinicians. The implications of this part of the research are that more research is needed on:

- which families with a child with CP need referral to more appropriate services
- how CDCs perceive their services (e.g. child centred or family centred)
- how CDCs are equipped to assess and form a family plan and make referrals.

One of the implications of the findings of this study was that many physiotherapists and allied professionals, who provide services for families of children with CP, need to re-examine their beliefs about the efficacy of physical therapy and the additional support they give to families. A recommendation is that therapists should receive in-service training on how to explain to parents that a very high intensity of physical therapies may not be warranted and how to assess families' requirements for other services.

The cost analysis suggested that increased funds may not be required, but that careful consideration should be given to the individual needs of the child and the family, with appropriate matching of such needs with service provision.

Finally, in this study various methodologies were used. The reviewers recommend that other assessments of therapies of this type adopt a similar multifaceted approach. This would seem to be more appropriate than a simple RCT for the evaluation of clinical interventions where the effects are complex.

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Contribution of authors

Michael Weindling (Professor of Perinatal Medicine), Sheila Glenn (Professor of Applied Developmental Psychology), Cliff Cunningham (Professor of Applied Psychology) and Rhiannon Edwards (Senior Research Fellow in Health Economics) designed the study and were responsible for its conduct, analysis and interpretation of the data. Professor Cunningham supervised the research assistants during the later part of the study. Dr Edwards was responsible for the health economics assessment. David Reeves (Senior Statistician) devised and carried out the statistical analysis. Professor Weindling was the principal investigator.

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Appendix 1

Selection, reliability and validity of measures

Pre-intervention assessments (T1) were available for 85 families on the child measures and 84 on the family measures. Unless stated otherwise, these data sets were used to examine the reliability and validity of the measures.

The response rates for specific variables vary due to some missing data. Response rates were given for T2, which included all 76 families who completed the intervention period and on whom we had pre- and post-intervention assessments.

Correlations were presented between T1 and T2 for these families and provide some indication of stability and reliability of the measures. All significance levels were for two-tailed tests and, unless stated otherwise, were at the 0.000 level.

Child outcome variables 1.1

The Gross Motor Function Measure[49–54]

This was developed in the late 1980s to assess the maturation of gross motor functioning up to the normal milestones of 5 years and at the same time be sensitive to the specific deficits resulting from CP.[49] It was the first standardised outcome measure designed for this purpose. There were 88 items divided into five dimensions: lying and rolling; sitting; crawling and kneeling; standing; and walking, running and jumping. Each item was scored on a four-point scale from the child 'does initiate the movement' to the child shows 'successful completion of the movement'. It was scored in terms of percentage of achievement of the items in the section and these can be summed and averaged to give an overall score. It is not a normative test and therefore does not indicate rate of development and degree of developmental delay. The few studies available have reported that the GMFM is a useful outcome measure of change in gross motor functioning of young children with CP, and have demonstrated reliability and validity (Olson DH: personal communication, 1995).[46–50,80]

Response rates T1 = 85; T2 = 76.

Reliability Inter-rater reliabilities were conducted on 15 joint assessments. The results of these indicated agreement between assessors of between 94 and 100% for each of the subscales. The T1–T2 correlation was 0.93, which was an indication of the reliability in terms on the rating of the children over the 6-month period.

Validity As expected, the GMFM correlated positively with the other measures of child functioning: Griffiths locomotor raw score (0.83), MQ (0.70), CQ (0.58), Vineland DL (0.68) and Vineland Socialisation (0.69).

The Griffiths Mental Development Scales[55]

These scales were devised in the 1970s for children from birth to 8 years of age. They were widely used by practitioners in the UK. There are six domains: locomotor; personal–social; hearing and speech; eye–hand coordination; performance; and, from the third year of life, practical reasoning. The scales have demonstrated reliability and validity from many studies. This was a normative test and the standardisation provided norms for developmental rate (age equivalent quotients), which could indicate the degree of developmental delay.

For the purposes of the current study, the raw scores were used. A total age equivalent score [developmental Quotient (DQ)] can be obtained but because of the motor impairment experienced by young children with CP and the requirement of motor skills on many of the performance items (e.g. block building), they may underscore on the total score of these developmental tests. The two domains with the minimum motor requirement are the personal–social and hearing and speech. The average of the age equivalent indexes for these two scales was used to compute a CQ. Similarly, the average for the locomotor and the eye–hand coordination domains was computed to form an MQ. These two quotients provided a normative measure of cognitive and motor functioning and so indicate level of impairment or degree of disability. The raw locomotor score was used as a comparison with the GMFM as both measure similar skills.

Response rate T1 = 85; T2 = 76.

Reliability Inter-rater reliabilities on 15 joint assessments resulted in agreements of above 95%

on each of the domain subscales. The T1–T2 correlation was 0.94.

Validity As stated above, there were significant correlations between the locomotor raw score and the GMFM score.

Motor quotient (MQ)

The correlations between the MQ and the GMFM, the Vineland DL and the Socialisation scores were 0.70, 0.37 and 0.45, respectively.

Cognitive quotient (CQ)

The correlations between the CQ and the GMFM, the Vineland DL and the Socialisation were 0.58, 0.46 and 0.56, respectively.

The order and pattern of these correlations were as expected. The CQ correlated 0.65 with the MQ.

The Vineland Adaptive Behaviour Scales[56]

This scale of adaptive functioning has been used in one form or another for over 50 years. The latest standardisation used in this study was in 1984. It comprises 577 items that were presented and scored by an interviewer based on caretaker responses. Around 100–150 items were appropriate for children aged under 4 years. There were four subscales: Communication, Daily Living (DL), Socialisation and Motor Skills. Each subscale produces a raw score that can be summed for a composite score. Standard scores and age equivalence scores can also be derived. In the present study, only the DL and the Socialisation scales raw scores were used. The contents of the remaining scales were assessed via the other child scales.

Response rates T1 = 85, T2 = 76.

Reliability The T1–T2 correlation was 0.90 for DL and 0.88 for Socialisation.

Validity As noted above, the expected pattern of positive correlations with the other child measures was found.

Family Outcome Measures

Parenting Stress Index (PSI)[81,82]

This measure is increasingly used in studies of young families with children with disability. There were 101 items rated on a five-point scale from 'strongly agree' to 'strongly disagree'. The PSI taps several areas of parent attitudes, feelings and stresses in the parent–child relationship and has shown to relate to child, parent and family characteristics and to life events. It yields two main summary scores: the Parent Domain score, which indicates stress from parenting sources, and the Child Domain score which indicates stress associated with the parent's perception of the actual child.

The Parent Domain has seven subscales (depression, attachment, restriction in role, sense of competence, social isolation, relationship with spouse/partner, parent health). This domain served as an outcome measure in the study.

The Child Domain has six subscales (adaptability, demandingness, mood, acceptability, reinforces parent, distractibility/hyperactivity). In the study by Shonkoff and colleagues in 1992,[27] the first three subscales were used to construct a measure of Child Temperament and found to be informative in the analysis of intervention effects and family functioning. In the present study, the correlation between Child Temperament and the total Child Domain score was 0.94 and the pattern of correlations with other variables was the same for both measures. Therefore, the Child Domain score and not the derived Child Temperament score was used in these analyses.

There was also a Life Event Stressors Scale. This measures and weights the events experienced during the months before completing the scale as potential stressors. It can therefore be used as a covariant when seeking to delineate the impact of other factors on the domain scores.

A number of items in the scale were used to check Defensive Responding. These have been compared to the Marlowe–Crowne Social Desirability Scale discussed later.

Threshold values over which one is advised to seek clinical assessment were provided for the domains. These were the total domain scores obtained by 85% or more of the sample. The manual provides scores for the 'normal' standardisation sample at different age groups. This is because the total stress score is associated with the age or developmental level of child. The manual also provides such scores for different populations, including CP. Hence a current sample can be compared with these existing data sets (see Appendix 2).

Missing data There were more missing data on this scale than expected. This issue has received little

attention in the previous literature, most studies merely stating that they used the criteria set out in the manual. On this basis, we would have lost around 30% of the data.

The problem was dealt with in three stages. First, an item analysis revealed several items with high levels of missing data. Inspection of these cases indicated that the item was inappropriate for this population or the parent. On the Parent Domain, five items (82–85, 87) referred to spouses and were not completed by the single parents. On the Child Domain, three items referred to physical movement of the child or assumed rapid developmental change (6, 22, 49). In these cases, the item was scored as a 5 ('highly disagree'). Second, the remaining missing items were dealt with according to the manual: the means were used to replace missing responses if five or fewer items were missing from the total scale or no more than two from any subscale. Following this, there were still substantial numbers of returned questionnaires that were rejected. The third stage began by determining the frequency of missing items in the 'rejected' questionnaires. This indicated that most had between five and 10 missing items evenly distributed across the two domains. It was decided to set a criterion of 10 items, the rationale being that the domain scores, not subscale scores, were going to be used in the analyses, and that the scores were to be used for research rather than clinical purposes, that is, group scores were to be used. Statistical advice and examination of the data confirmed this to be reasonable.

Response rate After controlling for missing data the response rate for each of the domain scores at T1 was 81 and at T2 it was 74.

Reliability Cronbach's alpha for the total scale was 0.94. Cronbach's alpha for the Parent Domain was 0.90 and for the Child Domain 0.91. The T1–T2 correlations were 0.81 for both Parent and Child Domains.

Validity For the Parent Domain there were positive correlations with Child Domain, Family Needs and GHQ of 0.62, 0.56 and 0.62, respectively, and with Maladaptive Coping (0.26, $p = 0.02$) and LOC (0.29, $p = 0.009$) and Life Event Stressors (0.21, $p = 0.053$). There were negative correlations with Family Cohesion (–0.39), Family Adaptability (–0.42) and both Formal Family Support (–0.28, $p = 0.011$) and Informal Family Support (–0.24, $p=0.035$). Hence higher stress scores on the Parent Domain were associated with greater family needs, more difficult children, lower mental health, less family cohesion and adaptability and maladaptive or passive coping mechanisms and lower internal LOC and family support. This was as expected from the studies on stress and coping and family functioning.

Similarly, for the Child Domain the significant correlations were with Family Needs, GHQ (0.33, $p = 0.002$), Family Adaptability (–0.23, $p = 0.04$), LOC (0.29, $p = 0.009$), Severity of Motor Impairment (–0.25, $p = 0.025$) and CQ (–0.38, $p = 0.001$).

The Life Events Stressor score was only correlated with Family Needs and, at the 10% level, the Parent Domain (PSI) (0.21, $p = 0.053$) and GHQ (0.20, $p = 0.06$). Thus, increased Life Event Stressors were associated with more expressed needs and slightly higher stress as indicated on the PSI and GHQ. The T1–T2 correlation was 0.55, which indicates the transient nature of some of these life events (also see comment on the GHQ below).

The General Health Questionnaire (GHQ)[59]

This was designed to detect psychiatric disorder in community settings and provides an index of mental health (feelings of anxiety, depression and social isolation). It has been widely used in research both to identify at-risk individuals and to measure degree of disorder. It was regarded as the best validated self-administered measure of this type for a British population. It has 12 items and respondents complete a four-point Likert scale (0–3) ranging from feeling 'better than usual' to 'much less than usual'. A second system of scoring provides thresholds of risk, indicating the need for further clinical appraisal. For this the items were scored as 0 or 1 and the thresholds were between 2 and 4 depending on the criteria set. This allows comparison between the current sample and other data sets (see Appendix 2). For the purposes of the validation exercise, we used the full score.

Missing data Missing data were not a problem for this scale. In the one instance where an item was missing, the mean value was imputed.

Response Rate After controlling for missing data there were 84 responses at T1 and 76 at T2.

Reliability Cronbach's alpha for the scale was 0.89. The T1–T2 correlation was 0.44, which, although highly significant, indicates relatively low stability. Given the scale's sensitivity to immediate stressful

events and circumstances, this might be expected. Life Events Stressors also shows relative low stability and supports this inference. Examination of the mean scores at T1 and T2 shows a small reduction of four points with similar ranges between the two time points. This again suggests individual variation.

Validity The expected positive correlation with the Parental Stress Index was noted above. Similarly, it was correlated with Family Needs, Maladaptive Coping (0.24, $p = 0.03$) and the Child Domain (0.33, $p = 0.002$). It was negatively associated with Family Cohesion (–0.32, $p = 0.003$), and Family Adaptability (–0.32, $p = 0.003$).

Family Needs Scale (FNS)[60]

An FNS was adapted from Bailey and Simeonsson.[60] There were 34 items and parents rated these on a three-point scale in relation to their child's disability or behaviour (definitely not needed, unsure, definitely needed). Higher scores indicate more needs. There were six subsections: needs for information; needs for support; help in explaining about their child to siblings, family, neighbours, etc.; help in obtaining community services; financial needs; and help with family functioning. In a previous study with families of children with complex needs, the scale was used in face-to-face interviews and then adjusted and turned into a postal questionnaire. The returns indicated that most families found it easy to use and did not suggest areas that were not covered. It was concluded the scale covered the whole range of needs.

Missing data Sixteen participants missed item 16. On inspection, this pertained to siblings and was not applicable to first-born children. Many mothers of first-borns had scored 1, not needed, so all the missing dated were coded the same. The few additional missing items were dispersed across the scale and were also coded as 1.

Response rate T1 = 83, T2 = 76.

Validity As noted above, the scale was correlated with the PSI and the GHQ. Positive correlations were found with the LOC (0.3, $p = 0.004$), Life Event Stressors (0.42) and the Child Domain score (0.42). There were negative correlations with Family Cohesion (–0.3, $p = 0.005$). Hence increased needs were associated with dysfunction and stress in families and lower internal LOC. Active coping mechanisms and family support could be expected to reduce family needs; however, no significant correlations were found with these measures. However, the subscale of 'need help with family functioning' negatively correlates with both Family Cohesion (–0.43) and Family Adaptability (–0.37). Hence, in terms of the expected association between stress and needs, and the family relationships, the scale has validity. The lack of strong correlation with the social support or coping scales suggests they may be measuring different factors and worth further exploration.

Parent satisfaction

Satisfaction was obtained from the parents in the PAG and the FSWG. Rather than use an existing scale, it was argued that an active interview procedure based on a structured protocol would allow a greater exploration of what parents liked/found helpful and did not like/find helpful about the intervention. The disadvantage was that the interviewer would become aware of the group and so independence could be compromised. To overcome this, parents were first asked to give a mark out of 10 for their overall satisfaction with the intervention, 1 being totally dissatisfied and 10 bring totally satisfied. About half of the interviews were conducted as part of the post-assessment by the independent assessor after completing the T2 assessments. They were completed shortly after the assessment by telephone by the project coordinator. Comparison of the means and range of the satisfaction ratings taken from the independent assessor interview with those of the project coordinator revealed no difference.

Missing data Due to a misinterpretation of the protocol, 17 parents were not asked to provide a 1–10 rating. The notes from the interviews were therefore used to rate these. To check for reliability, eight (21%) of the scored interviews were rated blind to knowledge of the actual score attained. The agreement with the rated and actual score was 87.5%.

Mediating variables and other measures

Family Support Scale (FSS)[83]

This self-administered scale has been extensively used in early intervention studies to measure the variety of formal and informal support provided to the family. It consists of 18 items and uses a five-point Likert scale from 'not at all helpful' to 'extremely helpful' to measure the degree of perceived helpfulness that the caretaker attributes

to each source of support. There was also an option to score 'not applicable' (0). A total score was obtained by adding the score for each item. Higher scores indicate increased support. The developers reported good reliability and validity (internal consistency reliability 0.77, split half reliability 0.75 and test–retest reliability 0.75). The scale can be separated into Informal and Formal Support to indicate the source and relevance of sources of support. Considerable research has demonstrated a positive correlation between satisfactory social support and parental well-being.

Missing data There were few missing items on completed questionnaires. Where these occurred, the item was coded as 'not applicable' (0).

Response rate T1 = 84, T2 = 75.

Reliability Cronbach's alpha for the total FSS score was 0.69. The T1–T2 correlation for the total score was 0.55, which was lower than the test–retest 0.75 reported in the manual. However, since the retest in the present study was 6 months later, the correlation was still acceptable.

Validity The FSS displayed a negative correlation with the Parental Domain of the Stress Index (–0.28, $p = 0.01$) and positive correlations with Family Cohesion (0.3, $p = 0.008$) and Family Adaptability (0.24, $p = 0.029$). It also positively correlated with Active and Adaptive Coping (0.23, $p = 0.04$, 0.28, $p = 0.01$) and negatively with LOC at the 10% level. Hence the expected relationship with parenting stress and family functioning was found and those parents with good coping and internal LOC appeared to perceive they were better supported.

The validity of the scale was reinforced when the two subscales were examined. The correlation between the Formal Support and Informal Support scores was 0.43, indicating that, although related, they were measuring discrete aspects. Both were significantly negatively correlated with Parental Stress. However, the Formal Scale and not the Informal Scale was significantly correlated with the LOC, Active and Adaptive Coping, suggesting that more able copers with higher internal levels of control were more likely to receive (i.e. use and seek out) formal support. Conversely, Informal Support and not Formal Support was significantly correlated with Family Cohesion and Adaptability, suggesting good relationships between members and with extended family.

Coping strategies (COPE)[47]

COPE has 52 items with four items in each subscale. The subscales are active coping, planning, seeking instrumental support, seeking emotional support, suppression of competing activities, turning to religion (not used in this study), positive reinterpretation and growth, restraint coping, acceptance, focus on venting emotions, denial, mental disengagement and behavioural disengagement. The manual reports Cronbach's alpha scores above 0.6 for all the subscales except the 'mental disengagement' scale. Like all such coping measures, the factor structure has not always been robust. However, the subscales were grouped into three factors, Active Coping, Adaptive Coping and Maladaptive Coping. The data in the present study were collated into the three factors and used in the analyses.

Missing data There were few missing items and these were dispersed throughout the scale. The means for each subscale were imputed if no more than one item was missing from that subscale.

Response rate After controlling for missing items, T1 = 84 and T2 = 76.

Reliability Cronbach's alpha scores for Active, Adaptive and Maladaptive Coping were 0.89, 0.76 and 0.75, respectively. The T1–T2 correlations were 0.60, 0.52 and 0.46, respectively.

Validity Active Coping was positively correlated with Family Cohesion (0.33, $p = 0.003$), Family Adaptability (0.31, $p = 0.004$) and Formal Family Support (0.35, $p = 0.001$). Adaptive Coping also correlated with Formal Family Support (0.38). Maladaptive Coping was positively correlated with the stress measures, namely GHQ (0.24, $p = 0.03$) and the PSI Parent Domain (0.26, $p = 0.017$).

Hence the scale appeared to be reasonably reliable and valid in indicating coping styles of the parents, although the expected relationship with the next measure, LOC, did not emerge.

Brief Locus of Control Scale (BLCS)[48]

One way of viewing control is as an individual's generalised belief about the ability to control important outcomes. It has been argued that internal LOC is associated with more successful coping. The BLCS was developed from the Rotter measure.[84] It consists of six items rated on a five-point scale, three of which tap internality and three externality. The lower the score, the greater is internal control. Lumpkin[48] reported a Cronbach's alpha of 0.68, significant correlations

with life satisfaction, health and coping and a mean score of 3.94 ± 0.46. In an earlier study with families of children with Down syndrome, we found Cronbach's alpha scores of 0.49 for mothers and a mean of 3.34 ± 0.43. Significant correlations were found with stress, parenting, social support and life satisfaction.

Missing data In the one instance of an item missing, the mid-point score was imputed.

Response rate T1 = 84, T2 = 76.

Reliability Cronbach's alpha was 0.31 for the total scale and 0.57 for internal items. The T1–T2 correlation was 0.38 for the total and 0.45 for the three internal items. Hence the reliability was not as good as expected.

Validity A positive correlation was found with maternal education level (0.23, $p = 0.034$) and at the 10% level with paternal education. When combined, the correlation was 0.24 and was in line with many other studies reporting a significant and positive correlation between internality and higher educational level. Also similar to previous studies were negative correlations with Family Cohesion (–0.27, $p = 0.015$), Formal Support (–0.25, $p = 0.024$) and the HOME scale score (–0.24, $p = 0.03$), indicating that externality was associated with reduced cohesion, perceived receipt of services and quality of home environment for the child. The expected positive correlations with stress, namely, the GHQ and PSI Parent Domain, were also found (0.23, $p = 0.035$ and 0.29, $p = 0.009$, respectively.)

Although the internal consistency and reliability were not that strong and suggested caution, the scale appeared to be adequately valid and reliable in the context of the present set of analyses.

Home Observation for Measurement of the Environment (HOME)[45]

This provides a rating of the child-focused nature of the environment using both observation and a semi-structured interview with the main caretaker – usually the mother. It consists of 45 binary items organised into six subscales: mother's responsiveness to the child; use of restriction and punishment; physical qualities of the home; availability of play materials; maternal involvement; and variety of daily stimulation. The higher the scores, the greater is the quality of the environment. It was a well-established measure with frequently reported acceptable levels of reliability. It had also been reported to correlate with socio-economic status and later IQ and language measures in children. One of its major functions was to identify homes that were likely to impede social and cognitive development.

Missing data None.

Response rate T1 = 85, T2 = 76.

Validity There were positive associations between the HOME and the Griffiths locomotor score (0.31, $p = 0.007$), Cognitive Status Index (0.26, $p = 0.017$), and the Vineland Socialisation Scale at the 10% level. There was a negative correlation with the Child Domain (–0.30, $p = 0.007$). Hence it appears that the more able the child, the greater is the likelihood of a higher rating of home environment, and the more the child is perceived as difficult (Child Domain), the lower the HOME rating, which is expected given that part of the HOME is about mother–child interactions. A positive correlation was found with Active Coping (0.22, $p = 0.05$) and negative with LOC (–0.24, $p = 0.03$) and, at the 10% level, with Maladaptive Coping. This again was in line with previous studies, which suggest that passive coping skills were associated with less active and engaging home environments for the child. No correlation was found with social class. This may be because socio-economic status, rather than social class, was a more sensitive measure of social deprivation. Also, very few parents in the present study were considered to be notably socially deprived by the independent assessor.

Family Adaptability and Cohesion Evaluation Scales (FACES III)

Both family cohesion and adaptability have been shown to be highly associated with positive and healthy development in young children and with engagement with services.

FACES III was a self-administered questionnaire comprising 20 items that measured two factors, emotional family cohesion and adaptability within the family. Each item was scored on a five-point Likert scale, where 1 indicated low cohesion and adaptability and 5 indicated high cohesion and adaptability. It was developed from the 30-item FACES II and although it had less reliable psychometric properties (Olson DH: personal communication, 1995), it was felt to be adequate for the current study and had the advantage of being shorter.

Missing data Several families failed to complete the whole questionnaire or did not complete three or more items. Hence the data were excluded. In line with the manual, the means were imputed when two or less items were missing.

Response T1 = 82, T2 = 67.

Reliability Cronbach's alpha score was 0.72, which was respectable. The T1–T2 correlation for Family Cohesion was 0.67 and for Family Adaptability 0.61.

Validity The two factors correlated 0.68, indicating a considerable amount of shared variance. Both positively correlated with Active Coping (0.33, $p = 0.003$ and 0.31, $p = 0.004$) and Informal and Formal Family Support (0.30, $p = 0.006$, and 0.28, $p = 0.013$). Both correlated negatively with the 'stress' measures of GHQ (–0.32, $p = 0.003$ for both) and the PSI Parent Domain (–0.39 and –0.42) and with the MQ (–0.28, $p = 0.01$ and –0.24, $p = 0.029$). At the 10% level, there were negative correlations with the motor scores (GMFM and Griffith locomotor score) and cognitive status (CQ). The validity was also supported by the differential correlations between the two factors and other variables. Thus, Paternal Education was correlated with Family Cohesion (–0.26, $p = 0.019$) and only at the 10% level with Family Adaptability. Family Cohesion was negatively correlated with Family Needs (–0.30, $p = 0.005$) and LOC (–0.27, $p = 0.015$) and only Family Adaptability was correlated with the Child Domain (–0.23, $p = 0.039$) and more strongly than the cohesion score with the GMFM and Griffith locomotor scores. This suggested that father's education was positively associated with cohesion in these families who also had lower family needs than other families, but may still find the child's physical disability was a problem to their adaptive functioning. There was also a negative correlation (–0.3) between social class and family cohesion. Since social class was correlated with education level (0.66), this again forms a valid pattern of associations.

Marlowe–Crowne Social Desirability Scale – short form[57]

In order to guard against responses being given because they were regarded as the 'right' responses for our culture, the Marlow–Crowne Social Desirability Scale was used. This has reasonable internal consistency and construct validity; for example, Strahan and Gerbasi[57] reported an overall reliability coefficient of 0.49 for American college females and 0.62 for UK males. The 10-item version of the scale was used in the current study. These were binary (true–false) items, which were scored 1 and 2, with the higher score indicating a greater degree of socially desirable responding. This version of the scale was least affected by age and socio-economic status and was particularly recommended for situations where internal reliability was less important than practical issues such as brevity and respondent burden in longer surveys.

Missing data These occurred on three completed questionnaires and affected between one and three items. In these instances, the mean was imputed.

Response T1 = 83, T2 = 73.

Reliability Cronbach's alpha = 0.55.

Validity The correlation with the defensive responding subscale of the PSI was –0.42. A low score on the PSI indicates defensive responding and a high score on the Marlowe–Crowne scale indicates socially desirable responding. The two measures were therefore associated as expected. A score of 24 or less on the PSI subscale indicated that the defensive responding was so great that the results may be unreliable. Two mothers had scores of 24 and one mother scored 21. This suggested that defensive responding and social desirability were not a cause for concern in this sample.

Measures of services received by the families

A key issue in any intervention study is assessing what other services providing similar help were being used by the participants. The issue of the effects of such services on the child's functioning is of paramount importance in the area of evidenced-based treatments. Several different sources of data were therefore used to create three measures of other services:

- additional services
- number of contacts
- physical therapy sessions.

Additional services

This was an attempt to measure the range and intensity of services received by each family. The main data sources were:

- Parent diaries kept on a daily basis throughout the intervention period and recording any services received with respect to the child.

- Assessor's questionnaire completed at the time of the pre- and post-assessment (T1 and T2).
- Family support worker diaries recording what they did during the weekly visits, which often included references to other contacts with the family.
- Project coordinator's notes from the initial interview of services currently being received by parents.
- A parent questionnaire, which was sent out following the intervention to all families to examine instances of incomplete diaries.

Comparisons were made between all the data sources and none were counter-indicative, that is, they supported each other. Information was likely to be left out rather than false information included, that is, services received when they were not. Hence all services from any source were amalgamated. These indicated that the parent diaries and the assessor's questionnaire were the most complete and accurate data sources.

Coding the parent diaries

- An independent researcher who was not part of the original study and who was unaware of the hypotheses read all parent diaries. Notes were made of the contacts and services received. A definition of additional services was devised. These were those services judged to be therapeutic or educational such as Occupational Therapy, Physiotherapy, School for Parents, Portage or Bobath. The last three of these are as follows. School for Parents (SfP) is a local initiative run by physiotherapy services from a CDC but providing a range of support including hydrotherapy, toy library, parent-to-parent and physiotherapy and occupational therapy. It is also open daily to families during school holidays. Portage is a home visiting early intervention programme for children with intellectual disability. It is usually weekly but sometimes less frequently and stops when a child enters nursery school full-time. Bobath is a private treatment which provides intensive physiotherapy and advice to parents over a short period of 1–2 weeks
- Visits to the hospital, the GP, the dentist and so on for mild health problems or check-ups were not included. The diaries were then re-read and, using this definition, placed into piles starting with no or few additional services up to lots of additional services. This indicated that all of the diaries could be accommodated within a seven-point scale. A second researcher repeated this exercise with a subset of diaries and there was over 90% agreement.
- From this, a code was devised to score the number and intensity of additional services:
  1 = nothing other than medical checks and visits from the research staff
  2 = occasional appointments less than once per month
  3 = occasional appointments once per month or more with one or two healthcare professionals
  4 = occasional appointments once per month or more with three or more health professionals
  5 = weekly appointments with one healthcare professional, possible additional appointments with other healthcare professionals
  6 = weekly appointments with at least two healthcare professionals, plus occasional appointments with others
  7 = intensive therapy with a range of healthcare professionals including Portage and/or Bobath.

This framework was used to score the 66 diaries. The scores ranged from 1 to 7 (mode 5.5, mean 5.07).

A second researcher independently coded seven diaries using the framework. Five codes were the same; the other two differed by one category (one lower and one higher) and both were mid-order ratings. Hence the coding was considered reliable.

Coding for the assessor's questionnaire

At the second assessment, parents were asked by the independent assessor what contact they had had with any health professional in the previous 6 months and how often this occurred. The questionnaire was completed for all 76 families at T2. The contacts included all hospital visits and visits to and by the GP and also physiotherapy, occupational therapy and so on. However, when compared with the other sources, it was clear that the responses focused on health professionals and did not record visits to the nursery or toy library. Also, the frequency was a broad rating recalled by the parent and did not appear to be very accurate when compared with the daily diaries.

The same coding scheme was used to rate the responses. The scores again ranged from 1 to 7 (mean 5.1).

Reliability The correlation between the scores from the parent diaries and the questionnaire was 0.56. The two sets of scores agreed in 45% of cases, and a further 38% were just one category different.

The differences were mainly accounted for by the parent failing to record visits with occupational therapy or speech therapy, or the assessor failing to record that the parent was receiving Portage home visiting or had taken part in an intensive private course of Bobath.

Where the scores agreed for both sets of data, this was taken as the final score. Where they differed, both sets of data were revisited. When necessary, the other relevant sources noted earlier were consulted. Because the differences were due to missed recordings of a service, any other noted service was added and used to compute the final rating. Hence all 76 participants received a code.

Number of contacts

These were computed from all the resources as noted above. This provided data on the number of contacts with services and on the visits received as a result of the intervention.

Physical therapy sessions

The Additional Services measure included regular attendance at a nursery or toddler group, which did not involve specific physical activities. Therefore, to explore the question of the impact of physical therapy activities on motor functioning, it was decided to create an index of physical therapy and obtain as strong a measure as possible of intensity.

The same procedure was used as described above. The measure encompassed all types of therapy that were based on physical contact. These included physiotherapy, hydrotherapy and occupational therapy. There was some ambiguity over specific services. It was decided to include Portage as many of the activities advised for these children included locomotor and eye–hand coordination tasks. After some deliberation, toy library was also included because many of the parents (particularly those in the Wirral using the SfP facility and some other CDCs which also had a toy library) noted a visit to the toy library; however, this included physiotherapy and listed the contact as someone who was known to be working as a physiotherapist. Visits for assessment only were not counted towards the measure.

It was not possible to arrive at an estimation of time spent in activities. We decided instead to use sessions as the unit measure. A session was calculated as each separate contact with the health professional, including the research PA or Portage home visitor. Thus, each home visit was a session. If the child saw both a physiotherapist and an occupational therapist at the child centre, this was calculated as two sessions. If a child attended a 2-week summer school, this would be recorded as 10 sessions of therapy, that is, morning and afternoon sessions 5 days per week for 2 weeks. If they saw the physiotherapist and then had hydrotherapy this also counted as two sessions, but if noted as hydrotherapy, this was one session. A large number of the children visited an SfP (see Box 1, p. 36) set up by a local paediatric physiotherapy service. Discussions with the staff gave us a clearer picture of the activities provided which was used to inform the ratings. The project coordinator, a senior paediatric physiotherapist who was aware of most service provision in the area, also advised.

Because parents recorded visits for various periods, the total number of physical therapy sessions was added together and divided by the number of weeks recorded. This resulted in a mean number of physical therapy sessions per week for the families completing the intervention period.

Two physical therapy sessions measures were available. Total physical therapy included all intervention visits, whereas physical therapy sessions excluded the intervention and gave an impression of ongoing services.

Independent variables

Parent education

Parents were asked when they had left school, if they had attended further education or training and, if yes, for how long and the qualifications obtained. With some exceptions, mothers gave the information for fathers. Two measures resulted:

- *Years in education:* age (a) up to 16, (b) 16 to 18 and (c) 18+ years (higher education). This was used in the randomisation procedure.
- *Educational qualifications:* five categories were used: (1) postgraduate; (2) graduate; (3) A-levels, BTEC, higher HNDs, currently studying; (4) GCSEs and/or some further training such as NVQ basic level; (5) no qualifications.

Social class

This was based on the Registrar General's Classification and parents were categorised based on their current or most recent occupation (*Table 22*).

As a result, 17% of the sample were classed as social class 5, 41% as social class 4, 40% as social

TABLE 22 *Social class*

Class	Examples of occupation in each class
1 Professional	Accountant, doctor, dentist, solicitor, university lecturer
2 Managerial and technical	Manager, teacher, librarian, nurse, farmer
3 (Non-manual) clerical and minor supervisory	Clerk, shop assistant, policeman, draughtsman, sales representative
3 (Manual) skilled manual	Electrician, tailor, bus driver, printer, cook
4 Semi-skilled manual	Agricultural worker, postman, fisherman, barman
5 Unskilled manual	Railway porter, labourer, lorry driver's mate, window cleaner, office cleaner

class 3, 9 % as social class 2 and 3% as social class 1. This skew was expected given the trend for higher incidence in social classes 4 and 5. The only significant correlations found were 0.66 with parental education and –0.3 with family cohesion.

Appendix 2

Levels of distress indicated by cases over thresholds on the Parent Stress Index and the General Health Questionnaire

Both the PSI and GHQ can be scored for threshold values indicating that the participant is at risk and should be referred on for further assessment (see Appendix 1).

Parent Stress Index

Comparison with typical population

The PSI manual provides total scores and domain scores (parent and child) for a range of percentiles. The 85th percentile level was selected as indicative of risk sufficient to warrant further investigation. The scores vary according to the age of the child (*Table 23*).

The present sample had a mean age of 19.9 months (SD 8.8) and so the 24-month scores were selected for comparison.

In the present sample, the percentage scoring above the thresholds were Total score 40%, Parent Domain 28% and Child Domain 43%.

Raw PSI scores are given in *Figure 12*.

The thick black line is the average PSI score for 32 children with CP used to derive the original test. The broken line is the average PSI score for 81 children with CP in the present sample. The similarity between the two patterns indicates the representability of the sample in this study compared with that of a sample of North American children with CP cited in the PSI manual.

TABLE 23 *PSI scores*

	Age (months)		
	12	**24**	**36**
Total	260	260	256
Child Domain	114	122	114
Parent Domain	150	149	143

Comparison with CP populations

A search of the literature found only one set of data using the PSI with a CP population and this was that quoted in the manual. It was based on the scores of 32 parents of children with CP aged 2 years (SD 1.1) and comparable to the present data based on 81 families with a young child with CP.

Inspection of the two profiles supports the conclusion that (a) families of young children with CP have considerably higher levels of stress as measured by the PSI and (b) the current sample compares closely with the previous sample supporting the notion of representation.

General Health Questionnaire

The GHQ has been used extensively as a measure of caseness (frequencies above/below threshold) in the UK.[55] The manual suggested using the score 2 as a threshold but most studies use 3, a score 4 or more being considered the most stringent threshold.

Using the T1 (N = 85) data set, 38% of mothers scored above 2, 32% above 3 and 27% above 4.

Comparison data for the general population were obtained from the Health Survey of England for 1993 and 1995 for women aged 16–44 years (i.e. covering the range of the present mothers). The threshold score was 4; 19.3% of 4318 cases scored above this threshold in 1993 and 21% of 4232 in 1995.

Using these criteria, the present score of 27% was higher. Because the present women were mothers of young children, one might have expected a higher score but, since the most stringent cut-off was used, the comparison supports the contention (and the PSI data) that a large number of the present mothers with a child with CP (and many had additional intellectual disability) were experiencing higher than average levels of stress.

Percentile	RAW PSI SCORES																
	Child Domain (CD)							Parent Domain (PD)									
	DI	AD	RE	DE	MO	AC	CD subscales total	CO	IS	AT	HE	RO	DP	SP	PD subscales total	CD and PD subscales total	No. of life stressors
99+	36	38	18	31	18	21	145	45	22	22	21	32	36	28	188	320	27
95	33	33	15	25	14	18	130	40	20	18	19	29	30	26	169	294	20
90	31	31	14	24	13	17	122	37	18	17	17	26	27	23	153	267	17
85	29	30	12	22	12	16	122	35	17	16	16	24	26	22	149	260	14
A																	A
80	28	28	11			15	114	34	16	15	15	23	24	21	142	252	12
75	27			21	11		111	33	15	14	14	22	23	20	137	244	11
70		27		20		14	108	32	14		13	21	22	19	132	239	10
65	26	26	10	19			105	31		13	12	20		18	129	234	9
60	25				10	13	102	30	13				21		126	228	8
55	24	25	9	18			100	29				19		17	123	224	7
50		24			9	12	99		12	12	11		20	16	121	222	6
45	23		8	17			97	28				18	19		118	217	
40		23				11	95	27				17		15	115	214	5
35	22	22		16	8		93	29	11	11	10		18	14	112	208	4
30			7			10	89	25				16	17		110	201	
25	21	21		15	7		87	24	10	10		15		13	107	195	3
20	20	20		14		9	82	23			9	14	16	12	102	188	2
15	19	19	6	13	6	8	78	22	9	9		13	15	11	99	180	
10	18	17		12		7	75	21	8	8	8	12	13	10	92	170	1
5	16	15					66	18	7		7	11	12	8	82	159	
1	9	11	5	9	5		50	15	6	7	5	8	9	7	69	131	

	DI	AD	RE	DE	MO	AC	Sub-total	CO	IS	AT	HE	RO	DP	SP	Sub-total	Overall total
North American reference group*	26	30	9	25	12	18	120	33	15	13	13	21	21	20	136	256
Present sample†	26	30	10	23	11	16	116	33	16	13	15	22	21	19	139	255

*Indicated by thick black line — Profile of mean scores for parents and children with CP in reference group ($N = 32$). Mean (± SD) age of the children was 24.0 ± 1.1 months.

†Indicated by broken line — Profile of mean scores for present sample ($N = 81$). Mean (± SD) age of the children was 19.8 ± 0.8 months.

A·····A — Clinical threshold level

Child Domain		Parent Domain	
DI	Distractibility/Hyperactivity	CO	Competence
AD	Adaptability	IS	Isolation
RE	Reinforces Parent	AT	Attachment
DE	Demandingness	HE	Health
MO	Mood	RO	Role Restriction
AC	Acceptability	DP	Depression
		SP	Spouse

FIGURE 12 *Raw PSI scores*

Appendix 3

Case studies from the family support worker diaries

These three case studies were selected to illustrate the extremely complex situations in which some of the PA and FSW interventions were embedded. In none of the cases were the parents referred by their GP or other health professionals to other services. In some cases this was due to the poor relationship with the health professionals and sometimes they were not seen as relevant to the problem, that is, they were only concerned with the child and his or her CP. We estimate that between 10 and 15% of the FSW sample experienced such problems and there was no reason to assume that the levels were different in the other two groups, given the randomisation.

Case study 1

This family consisted of a mother and father plus two children living in a family home. The father was educated to the age of 16 years and the mother to 18 years. Both were car driver/owners and worked in a family business. The child was diagnosed at 30 months of age as having spastic diplegia. She attended nursery three mornings per week.

From the beginning of the project, the mother disclosed to the FSW that she did not feel in control and that she was having problems with her husband, who kept taking himself off for the day and lying about it. This behaviour was a recurring issue throughout the diary. The mother clearly felt the need to talk about it with the FSW. He had also stopped helping with the children. Other family problems included the mother not speaking to the mother-in-law for a period of at least 3 months, while other members of the family made derogatory comments about how much input the family got from health professionals – implying that she was getting a lot of help and should be grateful. Despite this, the mother felt that she was having difficulties with the health professionals. These included the occupational therapist, who appeared to prefer to talk about other matters such as "tennis", rather than about the child, and the physiotherapist not turning up when an appointment was made. The mother was worried about the child with CP as she kept falling over. She also worried about the other sibling, who was currently experiencing health problems and had had a bowel operation previously. There were difficulties finding someone to child-mind for the sibling when the mother wanted to take the child with CP horse riding.

In the second month of the study, the father was laid off work. By the fourth month, the mother revealed that the family business was going under. As a consequence, she had to take a pay cut. Then her hours were cut. By the last month of the study, the mother admitted that she did not mix any more with other mothers at school or with friends. She had feelings of desperation when she just wanted to end it all. The FSW provided listening support and agreed to remain in contact at the end of the project.

Case study 2

This was a family of mother, father and two children. Both parents were educated up to age 18 years. The father was a car driver/owner, the mother was not. The child was diagnosed as having a hemiplegia at 11 months of age. At the start of the study, the family attended the SfP.

One month before the FSWs started to visit, the mother and father split up. According to the FSW diary, this was largely because the father could not face up to his responsibilities. He had also had children by other women and had left them. When he stopped paying maintenance, the mother sought legal advice. Despite this, they got back together again but split up three more times during the study period. In month four, they arranged a holiday together and decided to get married again at the father's suggestion, although they were not actually divorced. One month later, the father moved out again, saying that he did not love the mother. The mother then found out that she was pregnant but decided not to keep the baby as her marriage was over. The last entry described how the family were upset when the father did not appear on his birthday to spend it with the children until after they had gone to bed.

Other problems charted in the diary included the maternal grandmother being an alcoholic who

also suffered from angina and an aunt who always introduced the child as "the child with cerebral palsy", which upset the mother. In addition, the bailiffs once came round to the house because the family had not paid their council tax. There were no details in the diary illustrating how and if this was resolved.

The diary ended with the mother saying that the FSW had been a good support through the times that she had split from her husband. However, the FSW felt that she had let the mother down as she had to withdraw "at such a low point in the mother's life".

Case study 3

This was a two-parent family with three children. The youngest child was aged 8 months when diagnosed with mild tetraplegia. The mother was educated to age 18 years and was a car owner/driver, as was the father. His education level was not recorded.

At the start of the project, the mother was on Prozac, had had to give up her job and felt isolated. One reason noted for this was that when the child with CP was younger he had screamed all the time and family and friends had stopped visiting. The father told the FSW that he had not yet accepted the child as having CP as he could not see anything wrong with him. The mother was upset when he did not go with her to see the consultant. Towards the end of the project, the parents were not speaking to each other. This appeared to be due to the father believing that "the son is the mother's concern". She also reported that he spent time out of the house, while she feels like "a prisoner in her own home". By month six, the parents had separated.

During the project, other family problems emerged. One incident related to the eldest child being unaware that the father was not her biological father until she accidentally overheard it in a conversation. The mother also had health problems. In the 6-month study period she had a pregnancy scare, a lump in her throat, later diagnosed as a cyst, and a breast lump, for which she was told that she would have to have a biopsy (there was no further mention of this in the diary).

The diary also noted the mother's concerns with the child: problems with his feet, his gait and his walking. He also had behavioural problems, which included lashing out, biting and kicking.

In addition, the mother felt unsupported by some of the health professionals. She felt "betrayed" by the physiotherapist because of a tactless remark made by her and she had "no faith" in the health visitor, who had not been in touch.

Appendix 4

Steps taken to improve recruitment

Over the first 6 months only 17 children were recruited and by the end of the first year only 21. This was around 30–40% of the original estimates. Several initiatives were undertaken:

1. Letters were sent to the paediatricians and senior physiotherapists at the recruitment sites who had agreed to refer.
2. A study day was arranged (June 1998) and 65 people attended, including physiotherapists, occupational therapists and paediatricians. Further study days were held in September 1999, October 2000 and October 2001. They were all well attended and there was a feeling of support for the project.
3. The catchment area was increased and the age of the children was raised from under 3 to under 4 years.
4. Posters were also placed in CDCs and in Scope nurseries for families to self-refer.

In spite of these efforts, the recruitment rate remained at around 20 children each year.

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Feedback

The HTA Programme and the authors would like to know your views about this report.

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We look forward to hearing from you.

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