National Institute for Health and Care Excellence

Draft for consultation

Cerebral palsy in adults

[F1] Configuration of services: service design

NICE guideline tbc

Evidence reviews

July 2018

Draft for Consultation

These evidence reviews were developed by the National Guideline Alliance hosted by the Royal College of Obstetricians and Gynaecologists

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Contents

Configuration of services for adults with cerebral palsy who are over the age of 25 years

Review question

F1 What is the most clinical and cost effective configuration of services (setting and staffing) for adults with cerebral palsy?

Introduction

Children with cerebral palsy have access to services specifically related to cerebral palsy. When they become adults there is no such service available to them. To ensure that the adult's right for autonomy and independence are met they may need access to appropriate specialist services such as rehabilitation medicine, neurology, speech and language therapy services, physiotherapy and occupational health. Adults with learning disabilities (including adults with cerebral palsy who have learning disabilities) currently can have an annual review of their needs as outlined in the NICE guideline on challenging behaviour and learning disabilities. Adults with cerebral palsy, their family, or carers, may not be aware of their need for a specialist service, or know how to access that service. This review question looks at the evidence around the effectiveness of how these specialist services are accessed and delivered (including the effectiveness of the provision of an annual review to all adults with cerebral palsy).

PICO table

Please see Table 1 for a summary of the Population, Intervention, Comparison and Outcome (PICO) characteristics of this review.

Table 1: Summary of the protocol (PICO table)

Population	Adults aged 25 and over with cerebral palsy
Interventions	• Neurodisability focused services ○ MDT (core and extended combinations may include: speech and language therapist, occupational therapist, physiotherapist, psychologist, orthopaedic surgeon / neurosurgeon, neurologist, social worker, physiatrist (rehabilitation specialist), specialist nurse) ○ Service configurations used in other high income countries (for example: Canada, Holland and Sweden) • Learning disability services • Primary healthcare providers
Comparisons	• Other service configuration (as described in the study) • Each other
Outcome	**Critical** • Health-related quality of life • Time to treatment • Hospital admissions (unplanned) **Important** • Satisfaction (patient or carer reported) • Adverse effects (from delayed identification or management) • Residential care admissions (unplanned)

	- Length of hospital stay - Mortality

MDT: multidisciplinary team.

For full details see the review protocol in appendix A.

Methods and process

This evidence review was developed using the methods and process described in Developing NICE guidelines: the manual 2014. Methods specific to this review question are described in the review protocol in appendix A.

Declaration of interests were recorded according to NICE's 2014 conflicts of interest policy from May 2016 until April 2018. From April 2018 onwards they were recorded according to NICE's 2018 conflicts of interest policy. Those interests declared until April 2018 were reclassified according to NICE's 2018 conflicts of interest policy (see Interests Register).

Clinical evidence

Included studies

One retrospective cohort study (number of participants, N=345) was included in the review (Young 2005).

Young 2005 was a Canadian study of health service use in adults chronic and complex disorders originating in childhood, most of whom had cerebral palsy. The study reported accident and emergency department visits and hospital admissions in people whose outpatient care was provided by a single primary care physician versus those cared for by multiple doctors. In this study, care provided by a single primary care doctor was taken as an indicator of adequate health maintenance and promotion.

The clinical study included in this evidence review is summarised in Table 2 and evidence from this is summarised in the clinical evidence profiles below (Table 3).

See also the literature search strategy in appendix B, study selection flow chart in appendix C, forest plots in appendix E and study evidence tables in appendix D.

Excluded studies

Studies excluded from this systematic review, with reasons for their exclusion, are provided in appendix K.

Summary of clinical studies included in the evidence review

Table 2 provides a brief summary of the included study.

Table 2: Summary of included studies

Study	Design	Population	Comparison	Outcomes
Young 2005	Cohort study	345 adults (mean age 22 years) with chronic and complex physical disabilities of childhood. 58% had cerebral palsy, 25% spina bifida and 17% acquired brain injury in childhood. Canada	Outpatient care provided by a single primary care physician versus outpatient care provided by multiple different doctors.	- Hospital admissions (unplanned)

A&E: accident & emergency department.

See appendix D for the full evidence table.

Quality assessment of clinical outcomes included in the evidence review

The clinical evidence profile for this review question is presented in **Error! Reference source not found.**.

Table 3: Summary clinical evidence profile: primary healthcare providers versus other service configuration

Outcomes	**Illustrative comparative risks* (95% CI)**		**Relative effect (95% CI)**	**No of Participants (studies)**	**Quality of the evidence (GRADE)**
	Assumed risk with multiple different doctors	**Corresponding risk with single primary care doctor**			
HRQOL - not reported	-	-	-	-	-
Time to treatment - not reported	-	-	-	-	-
Visit to emergency dept. in past 4 years	583 per 1000	469 per 1000 (347 to 593)	OR 0.63 (0.38 to 1.04)	345 (1 study)	Very low[1,2,4]
Hospital admission in the last 4 years	314 per 1000	283 per 1000 (187 to 408)	OR 0.86 (0.5 to 1.5)	345 (1 study)	Very low[1,2,3,4]
Adverse effects - not reported	-	-	-	-	-
Unplanned residential care admissions - not reported	-	-	-	-	-
Length of hospital stay - not reported	-	-	-	-	-
Mortality - not reported	-	-	-	-	-
Satisfaction - not reported	-	-	-	-	-

CI: Confidence interval; HRQOL: health related quality of life; OR: Odds ratio;
1 Baseline differences in characteristics or severity of condition not controlled for in analysis
2 Only 58% of sample had cerebral palsy. Unclear whether the Canadian primary care model is applicable to the UK.
3 Unclear whether inpatient episodes were planned or unplanned.
4 Downgraded for imprecision: number of events < 300

See appendix F for the full GRADE table.

Economic evidence

Included studies

A systematic review of the economic literature was conducted but no studies were identified which were applicable to this review question.

Excluded studies

No studies were identified which were applicable to this review question.

Summary of studies included in the economic evidence review

No economic evaluations were included in this review.

Economic model

See appendix J for the full report of the economic model.

As no clinical evidence was identified as part of the clinical evidence review, threshold and "what-if" analyses were undertaken to identify the mean differences in unplanned hospital visits and length of hospital stay required for "ideal" service to be considered cost effective compared to a standard configuration of services. Costing was undertaken using a NHS and Personal Social Services (PSS) perspective. The model had a one year time horizon and consequently no discounting was undertaken.

Two outcomes (unplanned hospital visits [emergency department visits] and length of stay in hospital) specified in the protocol were explored in the model. 3 scenarios were considered to explore the differences needed for an "ideal" configuration of services to be cost effective:

1. Differences in emergency department visits excluding subsequent admissions to hospital
2. Differences in length of hospital stay excluding emergency department visits
3. Differences in emergency department visits followed by hospital admissions.

"Ideal" configuration consists of an annual 30 minute appointments with each of an adult physician, specialist nurse and physiotherapist or occupational therapist (90 minutes in total) as well as appointments with the individual's GP when and if needed. Standard configuration of services consists solely of when and if needed visits to the individual's GP with no other routine appointments.

A utility value of 0.513 identified from Dixon 2009 who elicited preferences from people in the UK using the EQ-5D 28 days following an emergency call compared to 0.80 in the baseline population was used to estimate a disutility of 0.0008 for a one day emergency room visit. Disutility associated with each day of stay in hospital was calculated from a weighted average from utilities reported for common reasons for hospital admissions also using the EQ-5D. Only the reasons for hospital admissions that are associated with cerebral palsy were used in the weighted estimate.

Costs for the model were taken from Curtis 2016 for healthcare professionals that care for adults with cerebral palsy. These costs are provided in Table 4, per working hour. Costs of unplanned hospital care are taken from NHS reference costs and are provided in Table 5Table 17.

Table 4: healthcare professionals providing care for adults with cerebral palsy

Health care professional	Cost
"Core" providing regular reviews	
Specialist nurse (band 6)	£44
Physiotherapist (band 6)	£45
Adult physician (consultant medical)	£135
"Extended" available as-and when-needed	
GP cost per hour of patient contact	£236
GP per surgery consultation 9.22 lasting minutes	£36
Consultant medical (neurologist, rehabilitation medicine or physiatrist)	£135

Health care professional	Cost
Consultant surgical (orthopaedic surgeon or neurosurgeon)	£137
Dietician (band 6)	£44
Speech and language therapist (band 6)	£44
Occupational therapist (band 6)	£45
Orthotist (band 6)	£45
Social worker	£79
Wheelchair services (NHS Reference Costs 2015/16, WC11 cost per review of all needs)	£152

NHS: National Health Service

Table 5: Cost of inpatient care

Reason for admission	NHS Reference Costs 2015/16 currency codes	Weight (Table 15)	Non-elective short stay cost applied to the first day of care [a]	Non-elective, excess bed day costs [a]
Pneumonia	DZ11K : DX11V	35%	£504	£259
Epilepsy	AA26C : AA26H	17%	£459	£297
Mental illness	SPHMSOTHAPC	15%	£429	£429
Lower, upper or other GI	FZ2G : FZ24J	33%	£923	£341
Weighted cost by reason	-	-	**£623**	**£317**

GI: gastrointestinal; NHS: National Health Service

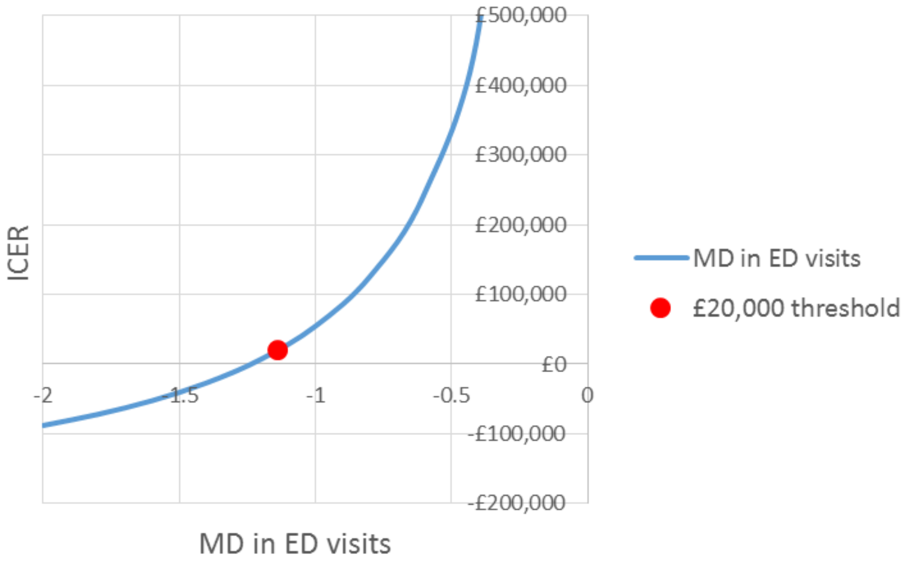
(a) Cost weighted by the Finished Consultant Episodes (FCEs) for each code

Results of the economic model

Emergency department visits excluding subsequent admissions to hospital

When a threshold analysis was conducted on the mean difference in emergency department visits “ideal” services would be considered cost effective at a threshold of £20,000 per QALY if the number of visits could be reduced by at least 1.14 per year. ICERs are illustrated in Figure 1 with mean differences ranging from 0 to -2 (no difference in visits to 2 less incurred by “ideal” services).

Figure 1: Threshold analysis for ED visits

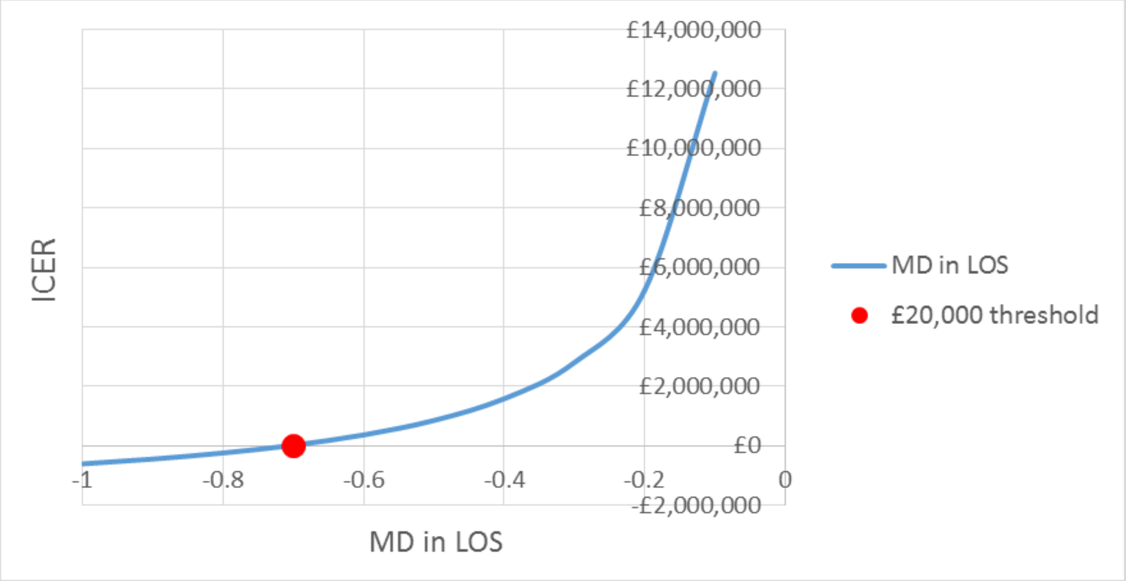


ED: emergency department; ICER: incremental cost effectiveness ratio; MD – mean difference

Admissions to inpatient care excluding emergency department visits

When a threshold analysis was conducted on the mean difference in the length of stay, “ideal” services would be considered cost effective at a threshold of £20,000 per QALY if the number of days could be reduced by at least 0.70. ICERs are illustrated in Figure 2Figure 11 with mean differences ranging from 0 to -1 (no difference in the length of stay to 1 less day incurred by “ideal” services).

Figure 2: Threshold analysis for LOS



ICER: incremental cost effectiveness ratio; LOS: length of hospital stay; MD: mean difference

Scenario analysis

Alternative scenarios considering the mean difference in emergency department visits or the length of hospital stay needed for “ideal” services to be the preferred option at a threshold of £20,000 per QALY are presented in Table 6. For example, a more frequent review (three times annually) would require a higher decrease in either emergency department visits (decrease of -3.42 visits) or length and stay (decrease of 2.1 days), compared to standard configuration of services, to be the preferred option. Less frequent or intensive reviews (e.g. 0.6 times per year or only conducted by the individual’s GP) would require less of a reduction in emergency department visits and/or to be the preferred option.

Table 6: Results of scenario analyses

Scenario [a]	MD in ED visits	MD in LOS
Base-case: reviews 1x year, 30 minutes in duration+30 minutes administration, conducted by an adult physician, specialist nurse and physiotherapist (or occupational therapist) compared with no routine reviews in standard care	-1.14	-0.70
Reviews as in base case but conducted 3x year	-3.42	-2.10
Reviews as in base case but conducted 0.6x year	-0.68	-0.42
Reviews as in base case but excluding an adult physician	-0.45	-0.28
Reviews 90 minutes (60 minutes+30 minutes administration) in duration	-1.71	-1.05
Standard services include 1 review with a GP lasting 18.44 minutes (lasting 2 standard consultation slots) plus a prescription cost [b]	-0.63	-0.39

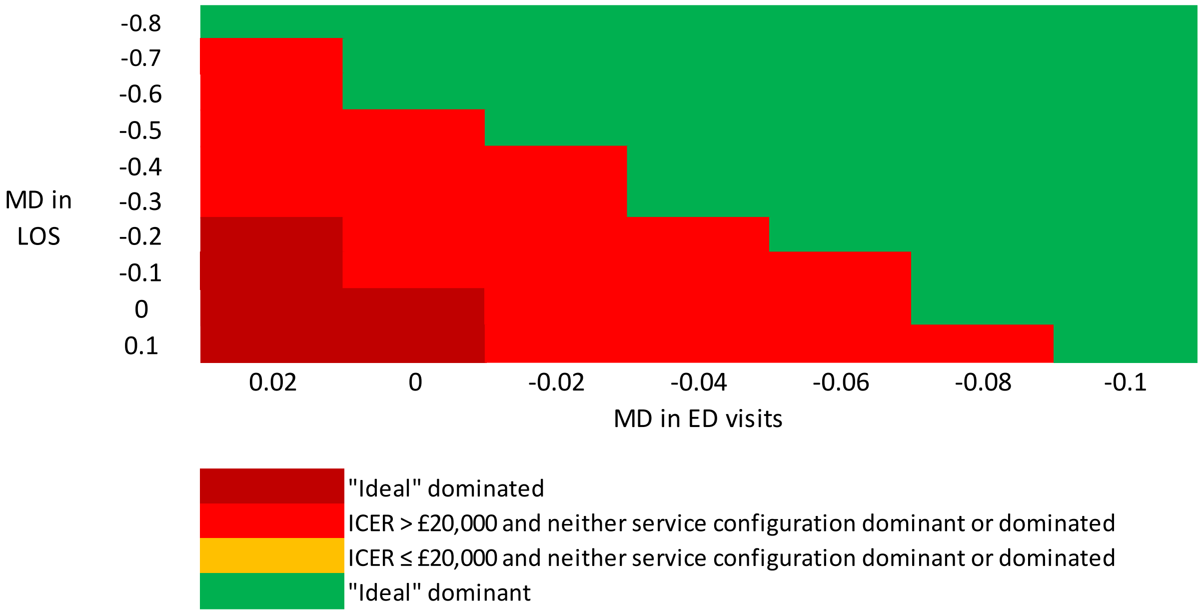
ED: Emergency Department; LOS: length of hospital stay; MD: m difference
(a) Holding all other inputs constant with base-case assumptions
(b) £28 prescription costs per consultation (PSSRU 2016)

Emergency department visits followed by a hospital stay

The results of varying the number of emergency department visits and length of hospital stay simultaneously are illustrated in Figure 3, using the base-case scenario. This analysis assumed that 100% of visits to the emergency department lead to an admission.

If we can assume "ideal" services reduced the length of stay by at least 0.7 days, "ideal" services could be considered cost effective compared to standard services, if "ideal" services do not increase the number of emergency department visits compared to standard services.

Figure 3: Threshold analysis varying the mean difference in emergency department visits and length of hospital stay



ED: Emergency department; ICER: incremental cost effectiveness ratio; LOS: length of hospital stay; MD: mean difference

Conclusions

Overall, only limited comparative clinical evidence was identified. This means that we cannot say with certainty which configuration of services will be cost effective, or what services or personnel should be included in the configuration. To address this uncertainty, the committee could consider subgroups of adults with cerebral palsy where specialist routine reviews are needed, as the benefits of a review will vary according to the person's needs and circumstances which may also change with time.

Evidence statements

Single primary healthcare provider versus other service configuration

Critical outcomes

Health-related quality of life

- No evidence was found for this outcome.

Time to treatment

- No evidence was found for this outcome.

Hospital admissions (unplanned)

- Very low quality evidence from 1 retrospective cohort study including 345 adults with complex physical disabilities of childhood suggested no clinically important difference in the rates of hospital admission when outpatient care was provided by single primary care doctor compared to multiple different doctors. It was unclear what proportion of these admissions were planned.
- Very low quality evidence from 1 retrospective cohort study including 345 adults with complex physical disabilities of childhood indicates that visits to the emergency department may be less likely when outpatient care was provided by single primary care doctor instead of multiple different doctors, but there was uncertainty about the effect estimate.

Important outcomes

Satisfaction (patient or carer reported)

- No evidence was found for this outcome.

Adverse effects (from delayed identification or management)

- No evidence was found for this outcome.

Residential care admissions (unplanned)

- No evidence was found for this outcome.

Length of hospital stay

- No evidence was found for this outcome.

Mortality

- No evidence was found for this outcome.

Recommendations

F1.1 For young adults moving from children’s to adults’ services, ensure that transitions to adult services are managed in line with the NICE guidelines on cerebral palsy in under 25s and transition from children’s to adults’ services for young people using health or social care services.

F1.2 Refer adults with cerebral palsy to a multidisciplinary team experienced in the management of neurological impairments if:

- Their ability to carry out their usual daily activities deteriorates **or**
- A neurosurgical or orthopaedic procedure is being considered that may affect their ability to carry out their usual daily activities

F1.3 Recognise that re-assessment by the multidisciplinary team and different clinical services may be needed by adults with cerebral palsy to ensure that their care meets their changing needs at different points in their lives.

F1.4 Commissioners and service providers should develop pathways that allow adults with cerebral palsy access to a local network of care that includes:

- advocacy support

- learning disability services
- mental health services
- orthopaedic surgery (and post-surgery rehabilitation)
- rehabilitation engineering services
- rehabilitation medicine or specialist neurology services
- secondary care expertise for managing comorbidities (for example, respiratory, gastrointestinal and urology services)
- social care
- specialist therapy services (for example physiotherapy and occupational therapy)
- wheelchair services.

F1.5 Ensure that adults with cerebral palsy, their families and carers, and their primary care teams are provided with information about their local network of specialist services.

F1.6 Explain to the person with cerebral palsy and their family members and carers their right to a care and support needs assessment, in line with the Care Act 2014, and discuss with them the type of support available.

F1.10 Consider regular reviews for adults with cerebral palsy, tailored to their needs and preferences. Agree with the person the frequency of review and which services should be involved based on their needs and preferences.

F1.11 Offer an annual review of the person's clinical and functional needs, carried out by a member of the clinical team, for people with cerebral palsy who have complex needs (such as GMFCS levels IV and V) and any of the following:

- with communication difficulties
- with learning disabilities
- in long-term care settings
- living in the community without sufficient practical and social support (for example, being cared for by elderly, frail parents)
- with multiple comorbidities.

F1.12 Discuss with the person with cerebral palsy (and their family and carers, if agreed) what information should inform the regular or annual review, and who should receive clinical information following review (for example, their GP).

F1.13 Record details of the person's review and share the information with relevant people (for example healthcare professionals and social care practitioners), with the person's permission.

F1.14 Discuss with the person with cerebral palsy (and their family and carers, if agreed) who will be their main point of contact between reviews.

F1.15 If an adult with cerebral palsy chooses not to have regular reviews, offer the person (and their family or carer, if agreed) information on when to contact a healthcare professional and how to access the specialist services that they may need. Ensure that the person's GP and multidisciplinary team are aware that they do not want to be reviewed regularly (with the person's permission).

F1.16 For adults with cerebral palsy and learning disabilities, offer an annual health check in primary care (see NHS England's information on annual health checks and NICE's guidelines on care and support of people growing older with learning disabilities and challenging behaviour and learning disabilities). Ensure that a referral to specialist services is made if a need for this is identified.

Rationale and impact

Why the committee made the recommendations

The committee discussed that specialist cerebral palsy services are provided for children and young people, but there is insufficient specialist service provision for adults, with variation and a lack of continuity in care. The committee highlighted that it is important that services continue to meet the people's needs when they move into adults' services, in line with recommendations in the NICE guidelines on cerebral palsy in under 25s and transition from children's to adults' services for young people using health or social care services. To ensure that needs are met for adults with cerebral palsy the committee made separate recommendations about service organisations (see below).

Disabled people have the same rights to access healthcare services as other people. These rights are outlined in the Equality Act 2010 and the UN Convention on the rights of persons with disabilities. To support this, the committee highlighted that there should be local pathways to enable access to a range of services for adults with cerebral palsy. The committee agreed that referral to specialist services is needed for some treatment options. Based on their discussions about treating spasticity and dystonia (see evidence reviews A1, A2 and A3), the committee agreed that access to specialist multidisciplinary teams is particularly important when a person with cerebral palsy experiences a deterioration in their ability to carry out usual daily activities, or when a neurosurgical or orthopaedic procedure is being considered that may affect their abilities. Adults with cerebral palsy may also need reassessment by the multidisciplinary team to take into account their changing needs at different life stages. The committee highlighted this and to raise awareness that people may need to access to different services if their needs change. These should include access to a multidisciplinary team if the person's functional abilities deteriorate or if the person's needs change.

The committee agreed that appropriate specialist services would need to be able to address a wide variety of impairments and comorbidities. To achieve good access to the relevant specialists, local networks of care are needed. The committee also noted that both people with cerebral palsy and healthcare professionals often lack awareness of the services available. Based on their experience, they agreed that provision of information about local networks of services would help to improve access.

The committee also acknowledged that carers are often unaware of their right to a local authority assessment of their own needs, including physical and mental health, and to an assessment of their need for respite care under the Care Act 2014. They agreed that highlighting this would help carers to access support if needed.

There was limited evidence for the effectiveness of different service configurations. One study focused on consistent outpatient care provided by a single doctor in primary care compared with care provided by several different doctors. Fewer visits to emergency departments and hospital visits were associated with having a single doctor. Although, the committee recognised that this may reflect the value of continuity of care, they agreed that the quality of the evidence was not good enough to make a recommendation.

Further evidence showed that changing the configuration of services to include the time and staff for an annual review, could be cost effective if there is a reduction of at least 1 emergency department visit per year per person. It was not clear from the evidence that this could be achieved when offered to all people with cerebral palsy. The committee discussed the variation in the needs of people with cerebral palsy and, based on their experience and knowledge, identified groups with more severe or complex health and social care needs who would most benefit from being offered an annual review. The evidence suggested annual review would be cost effective for this group because they decided that this would on average prevent one emergency department visit. It was noted that adults with learning disabilities should already be offered an annual health check in primary care.

Although the evidence did not support annual reviews for all people with cerebral palsy, the committee agreed that regular reviews are important to check for any new problems and ensure that people's needs continue to be met. They agreed that regular reviews should be considered, tailored to the person's needs. The frequency of review was not specified because it will depend on person's needs and wishes.

It is important to discuss who should be involved in the review with the adult with cerebral palsy. Information from different healthcare professionals and social care staff may be essential to fully understand the person's clinical needs. Good record keeping and sharing of information ensures that the outcome of the review is known to all relevant people and that the appropriate actions are taken. The committee also agreed that information about the review and any changes to the person's needs should be shared to ensure integrated care.

To help maintain continuity of care between reviews, the committee agreed that there should be a single point of contact, which could be a department or service in primary or secondary care.

The committee recognised that some people may not need or wish to have regular reviews. To ensure that they still have access specialist services when needed, the committee agreed that they should be given information on how and when to do this.

Impact of the recommendations on practice

The recommendations on access to services reinforce legislation and best current practice.

Service organisation for networks of care is variable, so changes to practice will depend on the availability of services within a particular local area. Providing information about local networks of care may incur an initial cost to set-up.

There may be an increase in the number of referrals. Training will also be needed to ensure healthcare professionals can manage referrals rapidly because there is a limited number of healthcare professionals with expertise in neurological disorders. However, recommendations with criteria for referral will help to reduce inappropriate referrals and variations in referrals.

Providing an annual review for adults with severe impairment will result in a minor change in practice and reduce variation.

Many adults with severe impairments will already receive an annual health check in accordance with the NICE guidelines on care and support of people growing older with learning disabilities and challenging behaviour and learning disabilities and NHS England's information on annual health checks.

There may be bigger changes in practice for people with less severe impairment if they are reviewed more often. However, the costs of this will be offset by the benefits of more frequent checks, such as early identification and management of new impairments or deterioration of function.

The committee's discussion of the evidence

Interpreting the evidence

The outcomes that matter most

The committee agreed that the critical outcomes for service configuration were health related quality of life, time to treatment and frequency and prevention of unplanned hospital admissions. This was because service configuration influences whether the diverse health needs of adults with cerebral palsy can be met in a planned way, avoiding emergency hospital admissions, treatment delays and poor health related quality of life. Other outcomes that the committee decided were important were satisfaction with the service (self or carer reported), unplanned residential care admissions, length of hospital stay and mortality rate. The committee chose these outcomes to reflect both the impact on the person with cerebral palsy as well as the implications that services have on costs.

In the clinical review only one study was identified which reported the outcomes 'visit to emergency department in past 4 years' and 'hospital admission in the last 4 years'. No other outcomes were reported.

The quality of the evidence

The quality of the outcomes reported in the identified study was very low according to GRADE standards. The intervention was indirect because there are many different reasons why care could be provided by a single, primary care physician rather than a number of different physicians. It was noted by the committee that there were baseline differences that indicated that people with a number of physicians were also those with more severe impairments. The study did not account for any possible confounding factors related to this or other factors that could affect outcomes 4 years later. The committee also discussed that less than two-thirds of people in this study had cerebral palsy which also limited the generalisability of the findings. In addition it was noted that the effect estimates had wide confidence intervals which made also impacted the confidence that the committee placed on these findings.

Overall the committee had limited confidence in the evidence and based their recommendations mainly on findings of the economic model as well as on their experience and expertise.

Benefits and harms

Even though the committee acknowledged the very serious limitations of the clinical evidence, they discussed that consistency and continuity of care is not always adequate in the current service provision for adults with cerebral palsy. Based on their experience and consensus, the committee agreed that the transition process from children's disability services to general adult services may have a detrimental effect on meeting the specific needs that adults with cerebral palsy have. These transition processes have been previously discussed in related NICE guidelines (cerebral palsy in under 25s and transition from children's to adults' services for young people using health or social care services) to which the committee cross-referred.

Based on their experience and expertise and taking into account of the management options for spasticity and dystonia (see evidence reviews A1, A2 and A3) that mean that a number of treatments could neither be prescribed or administered in primary care, the committee agreed that referral to specialist services is therefore necessary. Rapid access to specialist multidisciplinary teams is particularly important when the person with cerebral palsy experiences a change or deterioration in their functional abilities. The committee considered, based on their experience and expertise, a variety of different pathway models for clinical care for adults with cerebral palsy. They recognised that the needs of adults with cerebral palsy may change over time and that services should be sufficiently flexible to manage these challenges.

The committee discussed that there is wide variation in the needs of people with cerebral palsy and that they could therefore not be too prescriptive about how a specialist service is configured. However, they agreed based on their experience and expertise, that services are not always joined up to provide all aspects of care. To enable individualised care the committee recommended that pathways need to be developed to provide access to a range of services (for example mental health and movement therapy, including orthopaedic surgery). Such pathways, the committee noted, would lead to the more timely treatment and may prevent unplanned hospital admissions.

In order to empower them in their own clinical choices, information about these local networks should be shared with adults with cerebral palsy, their families, carers and primary care teams.

Services should also ensure that information is shared about the care and support needs assessment to ensure that the adult with cerebral palsy receives appropriate care and that their family and carers are sufficiently supported. The committee acknowledged that this is in line with the Care Act 2014 and reinforces this legislation.

The committee believed that regular reviews would be beneficial in ensuring continuity of care and reducing emergency hospital admissions. The committee discussed that there is wide variability in clinical needs that may also change over time. Although the evidence presented was not strong enough to recommend annual reviews for all people with cerebral palsy, the committee agreed that regular reviews, tailored to the person's needs, could be considered. The frequency of these reviews was not specified because it would depend on the individual's needs and preferences.

Based on their experience and knowledge they identified groups with more severe or complex health and social care needs who would most benefit from being offered an annual review. They noted that the additional resources would be outweighed by cost saving related to emergency department visits. This was also supported by the economic evidence.

The committee agreed that the review should be based on sufficient information and this could come from different sources. They discussed that potentially a range of healthcare professionals or social care practitioners can be in regular contact with the adult with cerebral palsy and may have useful knowledge to inform the review. The committee therefore recommended that a discussion should take place with the person to find out which information could be used as the basis for the review. For continuity of care it is also important to document the results of the review and share it with the relevant people in order to ensure that the appropriate actions are taken forward after the review.

Regardless of the type of service they therefore recommended that there should be an identified primary point of contact which could be a department or service in primary or secondary care to maintain continuity of care.

The committee recognised that some people may not need or wish to have regular reviews. To ensure that they still have access to specialist services when needed, the committee agreed that health and social care professionals should provide information on how the individual could do this. The committee noted that it would be important to share the information that the person decided not to have a review with other healthcare professionals with their agreement so that they are aware of this and ensure that the relevant support is provided regardless of whether or not they had a review.

It was noted that adults with learning disabilities should already be offered an annual health check in primary care and cross-references to the relevant guidelines were made In order to maximise the benefits of such a review they noted that referrals should be made if a need is identified during the review.

Cost effectiveness and resource use

The committee noted that no relevant published economic evaluations had been identified for this topic. They also agreed that it would not be cost effective to provide a full time multidisciplinary team for all adults with cerebral palsy, as people with mild impairments may not benefit from the additional care. On the other hand, those with severe impairments will need their care reviewed regularly to meet their changing needs. Therefore, to promote a cost effective use of resources, the committee considered the needs of adults in their recommendations to ensure the cost of a recommended service was offset by the health benefits it could provide.

The economic model developed for this review was used to assess the cost effectiveness of services in the absence of comparative clinical evidence. The committee agreed routine reviews could reduce the number of visits to the emergency department and explored the mean difference required for a number of configurations. The committee also considered the relationship between emergency department visits and subsequent hospital stays, but agreed it was difficult to model this relationship when the reasons for admission can depend on the needs of the individual and other factors.

Identifying a main point of contact promotes a transparent and efficient service at a negligible administration cost.

Following this, the committee agreed clinical services should develop pathways that allow access to a local network of care for the services regularly sought by adults with cerebral palsy. Developing those pathways may incur implementation costs where networks are unknown, but would promote timely access to the appropriate healthcare professional, leading to better identification and thus more timely management. As a result, some of the investment to identify those networks may reduce downstream costs.

The committee noted that the diagnosis and management of cerebral palsy is a core element of training in paediatrics and that paediatric patients are provided with a specialist MDT. In adult practice cerebral palsy is less likely to be included in training. The majority of adults with cerebral palsy are seen by their GP or general physician who may not have specialist expertise in cerebral palsy. Ideally the committee wanted adults with cerebral palsy to have access to healthcare professionals with a specialist interest in neurodisability, but acknowledged this may not be achievable without additional training or waiting lists.

In adulthood, primary care is usually where specific medical needs are most likely to be identified. However, when there is a change or deterioration in a person’s functional abilities, the committee agreed specialist advice should be sought to reduce delays in management or costly unplanned visits to hospital.

To reduce the number of unidentified or unmanaged problems, the committee made a recommendation to offer adults with cerebral palsy who have complex needs, multiple medical co-morbidities, or cognitive or communication impairments an annual review. This would focus on the assessment and management of their clinical and functional needs with an appropriate member of the multidisciplinary team. The cost of those routine reviews could be offset by fewer GP visits, fewer unplanned admissions to hospital and a higher quality of life as the committee believed many adults with severe cognitive or communication impairments live with those problems until they are unbearable, or recognised as an emergency.

Other factors the committee took into account

The committee noted that there are specific considerations to make in the transition from children’s to adult’s services. They therefore cross-referenced to existing NICE guidelines which addresses this topic, i.e. cerebral palsy in under 25s and transition from children’s to adults’ services for young people using health or social care services. The committee also noted that it is currently recommended that adults with learning disabilities receive and annual review and they therefore cross-referenced to the NICE guideline on challenging behaviour and learning disabilities as well as NHS England guidance (NHS England’s information on annual health checks) to ensure that such reviews are offered to adults with cerebral palsy who have learning disabilities.

Appendices

Appendix A – Review protocols

Review protocol for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

Table 7: Review protocol for organisation of services

[**Field (based on PRISMA-P)**	**Content**
Review question	What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?
Type of review question	Intervention
Objective of the review	The aim of this review is to determine whether particular service configurations are more clinically and cost effective than others.
Eligibility criteria – **population**/disease/condition/issue/domain	Adults aged 25 and over with cerebral palsy
Eligibility criteria – **intervention**(s)/exposure(s)/prognostic factor(s)	• Neurodisability focused services ○ MDT (core and extended combinations may include: speech and language therapist, occupational therapist, physiotherapist, psychologist, orthopaedic surgeon / neurosurgeon, neurologist, social worker, physiatrist (rehabilitation specialist), specialist nurse) ○ Service configurations used in other high income countries (for example: Canada, Holland and Sweden) • Learning disability services • Primary healthcare providers
Eligibility criteria – **comparator(s)/**control or reference (gold) standard	• Other service configuration (as described in the study) • Each other
Outcomes and prioritisation	**Critical** • Health-related quality of life • Time to treatment

[**Field (based on PRISMA-P)**	**Content**
	• Hospital admissions (unplanned) **Important** • Satisfaction (patient or carer reported) • Adverse effects (from delayed identification or management) • Residential care admissions (unplanned) • Length of hospital stay • Mortality Minimally important differences • Dichotomous outcomes will use default MIDs [RR thresholds of 0.80 and 1.2] • Continuous outcomes will use default MIDs [0.5 times the SD of the control group]
Eligibility criteria – **study design**	• RCTs (including conference abstracts of RCTs) • Comparative (prospective and retrospective) cohort studies • Cross-sectional studies • Registry and audit data
Other inclusion **exclusion criteria**	None
Proposed sensitivity/**sub-group analysis**, or meta-regression	Population subgroups: • GMFCS level • Learning disability • Communication ability Intervention subgroups: Adults with cerebral palsy living in residential homes • Process of transition from paediatric services
Selection process – duplicate screening/selection/analysis	A random sample of the references identified in the search will be sifted by a second reviewer. This sample size will be 10% of the total, or 100 studies if the search identifies fewer than 1000 studies. All disagreements in study inclusion will be discussed and resolved between the two reviewers. The

[Field (based on PRISMA-P)	Content
	senior systematic reviewer or guideline lead will be involved if discrepancies cannot be resolved between the two reviewers.
Data management (software)	STAR was used for study sifting
Information sources – databases and dates	Embase 1974 to present, Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R) 1946 to Present, PsycINFO 1806 to present
Identify if an update	Not an update
Author contacts	For details please see the guideline in development web site.
Highlight if amendment to previous protocol	For details please see section 4.5 of Developing NICE guidelines: the manual 2014
Search strategy – for one database	For details please see appendix B.
Data collection process – forms/duplicate	A standardised evidence table format will be used, and published as appendix D (clinical evidence tables) or H (economic evidence tables).
Data items – define all variables to be collected	For details please see evidence tables in appendix D (clinical evidence tables) or H (economic evidence tables).
Methods for assessing bias at outcome/study level	Standard study checklists were used to critically appraise individual studies. For details please see section 6.2 of Developing NICE guidelines: the manual 2014 The risk of bias across all available evidence was evaluated for each outcome using an adaptation of the 'Grading of Recommendations Assessment, Development and Evaluation (GRADE) toolbox' developed by the international GRADE working group http://www.gradeworkinggroup.org/
Criteria for quantitative synthesis	For details please see section 6.4 of Developing NICE guidelines: the manual 2014
Methods for quantitative analysis – combining studies and exploring (in)consistency	For details please see the methods in supplementary document C.
Meta-bias assessment – publication bias, selective reporting bias	For details please see section 6.2 of Developing NICE guidelines: the manual 2014
Confidence in cumulative evidence	For details please see sections 6.4 and 9.1 of Developing NICE guidelines: the manual 2014
Rationale/context – what is known	For details please see the introduction to the evidence review.
Describe contributions of authors and guarantor	A multidisciplinary committee developed the evidence review. The committee was convened by the National Guideline Alliance (NGA) and chaired by Dr Paul Eunson in line with section 3 of Developing NICE guidelines: the manual 2014.

[**Field (based on PRISMA-P)**	**Content**
	Staff from the NGA undertook systematic literature searches, appraised the evidence, conducted meta-analysis and cost effectiveness analysis where appropriate, and drafted the guideline in collaboration with the committee. For details please see the methods chapter of the full guideline.
Sources of funding/support	The NGA is funded by NICE and hosted by the Royal College of Obstetricians and Gynaecologists.
Name of sponsor	The NGA is funded by NICE and hosted by the Royal College of Obstetricians and Gynaecologists.
Roles of sponsor	NICE funds NGA to develop guidelines for those working in the NHS, public health and social care in England
PROSPERO registration number	Not applicable

GRADE: Grading of Recommendations Assessment, Development and Evaluation; GMFCS, gross motor function classification system; HTA: Health Technology Assessment; MDT: multidisciplinary team; MID: minimally important difference; NGA: National Guideline Alliance; NHS: National Health Service; NICE: National Institute for Health and Care Excellence; RCT: randomised controlled trial; RoB: risk of bias; SD: standard deviation

Appendix B – Literature search strategies

Literature search strategies for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

Database: Medline & Embase (Multifile)

Database(s): Embase 1974 to 2018 March 22, Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R) 1946 to Present, PsycINFO 1806 to March Week 3 2018

Table 8: Last searched on 22 March 2018

#	Searches
1	exp Cerebral Palsy/ use prmz
2	exp cerebral palsy/ use oemezd
3	exp Cerebral Palsy/ use psyh
4	((cerebral or brain or central) adj2 (pal* or paralys#s or pares#s)).tw.
5	cerebral palsy.ti,ab.
6	little? disease.tw.
7	((hemipleg* or dipleg* or tripleg* or quadripleg* or unilateral*) adj5 spastic*).tw.
8	((hemipleg* or dipleg* or tripleg* or quadripleg* or unilateral*) adj3 ataxi*).tw.
9	or/1-8
10	limit 9 to english language
11	limit 10 to (adult <18 to 64 years> or aged <65+ years>) use oemezd [Limit not valid in Ovid MEDLINE(R),Ovid MEDLINE(R) In-Process,PsycINFO; records were retained]
12	limit 10 to "all adult (19 plus years)" [Limit not valid in Embase,PsycINFO; records were retained]
13	12 use prmz
14	limit 10 to adulthood <18+ years> [Limit not valid in Embase,Ovid MEDLINE(R),Ovid MEDLINE(R) In-Process; records were retained]
15	14 use psyh
16	or/11,13,15
17	exp "delivery of health care"/ or exp health services administration/ or exp models, organizational/ or exp models, nursing/ or exp home health nursing/ or exp community health services/ or exp community health nursing/ or exp health services accessibility/ or exp Health Education/ or exp Health Care Rationing/ or exp Health Personnel/ or exp "Attitude of Health Personnel"/ or exp Patient Care Team/ or exp Health Status Indicators/ or exp Health Status/ or exp Rehabilitation Centers/ or exp "Physical and Rehabilitation Medicine"/ or exp Neurological Rehabilitation/ or exp Rehabilitation Nursing/ or exp Rehabilitation, Vocational/ or exp Rehabilitation/ or exp Psychiatric Rehabilitation/ or exp "Continuity of Patient Care"/ or exp Professional-Family Relations/ or exp Primary Health Care/ or exp "Patient Acceptance of Health Care"/ or exp "Quality of Health Care"/ or exp "Quality of Life"/ or exp Mental Health Services/ or exp "Health Services Needs and Demand"/ or exp Needs Assessment/ or exp Community Mental Health Services/ or exp Public Policy/ or exp Health Policy/ or exp "Standard of Care"/ or exp Professional Practice/ or exp Medical Audit/ or exp Clinical Audit/ or exp "Utilization Review"/ or exp State Medicine/ or exp Social Support/ or exp Social Work/ or exp Health Services for the Aged/ or exp Education, Special/ or exp Employment/ or exp Financing, Government/ or exp National Health Programs/ or exp "Referral and

#	Searches
	Consultation"/ or exp Hospitalization/ or exp Disability Evaluation/ or exp Disease Management/ or exp "Severity of Illness Index"/ or exp "Transportation of Patients"/ or exp Patient Preference/ or exp "Outcome and Process Assessment (Health Care)"/ or exp Sickness Impact Profile/ or exp "Activities of Daily Living"/ or exp Self Care/ or exp Exercise Therapy/ or exp Home Care Services/ or exp Complementary Therapies/ or exp Recreation/ or exp Leisure Activities/ or exp Age Factors/ or exp Life Style/ or exp Transition to Adult Care/ or exp Socioeconomic Factors/ or exp Aftercare/ or exp Speech Therapy/ or exp Language Therapy/ or exp Occupational Therapy/ or exp Physical Therapy Modalities/ or exp Behavior Therapy/ or exp Psychotherapy/ or exp Cognitive Therapy/ or exp Orthopedics/ or exp Neurosurgeons/ or exp Neurologists/ or exp Counseling/ or exp Counselors/ or exp Social Workers/ or exp Physiatrists/ or exp Psychiatry/ or exp Epidemiologists/ or exp Dietetics/ or exp Nutritionists/ or exp Neurodevelopmental Disorders/rh [Rehabilitation]
18	17 use prmz
19	exp health care delivery/ or exp health service/ or exp nonbiological model/ or exp model/ or exp home care/ or exp community care/ or exp community health nursing/ or exp health education/ or exp health care organization/ or exp health care personnel/ or exp health personnel attitude/ or exp patient care/ or exp health status indicator/ or exp health status/ or exp rehabilitation medicine/ or exp neurorehabilitation/ or exp rehabilitation center/ or exp rehabilitation/ or exp rehabilitation nursing/ or exp vocational rehabilitation/ or exp psychosocial rehabilitation/ or exp human relation/ or exp primary health care/ or exp patient attitude/ or exp health care quality/ or exp "quality of life"/ or exp mental health service/ or exp needs assessment/ or exp public policy/ or exp health care policy/ or exp professional practice/ or exp clinical audit/ or exp "utilization review"/ or exp national health service/ or exp social support/ or exp social work/ or exp elderly care/ or exp education/ or exp employment/ or exp financial management/ or exp public health/ or exp patient referral/ or exp hospitalization/ or exp disability/ or exp disease management/ or exp "severity of illness index"/ or exp patient transport/ or exp patient preference/ or exp treatment outcome/ or exp Sickness Impact Profile/ or exp daily life activity/ or exp self care/ or exp kinesiotherapy/ or exp alternative medicine/ or exp recreation/ or exp leisure/ or age/ or exp lifestyle/ or exp transition to adult care/ or exp socioeconomics/ or exp aftercare/ or exp speech therapy/ or exp language therapy/ or exp occupational therapy/ or exp physiotherapy/ or exp behavior therapy/ or exp psychotherapy/ or exp cognitive therapy/ or exp counseling/ or exp neurosurgeon/ or exp neurologist/ or exp social worker/ or exp physiatrist/ or exp orthopedic surgeon/ or exp doctor patient relation/ or exp counseling/ or counselor/ or exp psychiatrist/ or exp epidemiologist/ or exp dietitian/ or exp "organization and management"/ or exp teamwork/ or exp mental disease/rh [Rehabilitation]
20	19 use oemezd
21	(((exp Health Care Utilization/ or exp "Quality of Care"/ or exp Health Care Reform/ or exp Mental Health Services/ or exp Health Personnel Attitudes/ or exp Health Care Services/ or exp Health Care Delivery/ or exp Health/ or exp Health Care Policy/ or exp Models/ or exp Organizational Effectiveness/ or exp Organizational Structure/ or exp Organizational Development/ or exp Nursing/ or exp Home Visiting Programs/ or exp Home Care/ or exp Public Health Service Nurses/ or exp Hospitalization/ or exp Public Health/ or exp Public Health Services/ or exp Community Health/ or exp Community Mental Health Services/ or exp Community Services/ or exp Health Promotion/ or exp Health Service Needs/ or exp Health Education/ or exp Resource Allocation/ or exp Health Personnel/ or exp Interdisciplinary Treatment Approach/ or exp Work Teams/ or exp Teams/ or exp Health Disparities/ or exp Rehabilitation Centers/ or exp Rehabilitation/ or exp Neuropsychological Rehabilitation/ or exp Neurorehabilitation/ or exp Nursing/) and exp Rehabilitation/) or exp Vocational Rehabilitation/ or exp Mental Disorders/ or exp "Continuum of Care"/ or exp Primary Health Care/ or exp Client Attitudes/ or exp "Quality of Life"/ or exp Needs Assessment/ or exp Government Policy Making/ or exp Professional Standards/ or exp Clinical Practice/ or exp Clinical Audits/ or exp Utilization Reviews/ or exp Alternative

#	Searches
	Medicine/ or exp Social Support/ or exp Social Casework/ or exp Aging/ or exp Special Education/ or exp Employment Status/ or exp Professional Referral/ or exp Professional Consultation/ or exp Consultation Liaison Psychiatry/ or exp Disability Evaluation/ or exp Disease Management/ or exp "Severity (Disorders)"/ or exp Treatment Barriers/ or exp Preferences/ or exp Decision Making/ or exp "Activities of Daily Living"/ or exp Self-Care Skills/ or exp Self-Management/ or exp Self-Efficacy/ or exp Coping Behavior/ or exp Exercise/ or exp recreation/ or exp recreation therapy/ or exp Well Being/ or exp Leisure Time/ or exp Participation/ or exp Lifestyle/ or exp Client Transfer/ or exp Socioeconomic Status/ or exp aftercare/ or exp Language Therapy/ or exp Speech Therapy/ or exp Occupational Therapy/ or exp Physical Therapy/ or exp Psychotherapy/ or exp Behavior Therapy/ or exp Cognitive Therapy/ or exp Surgeons/ or exp neurologists/ or exp Social Workers/ or exp Neurology/ or exp Physicians/ or exp psychiatrists/ or exp psychiatry/ or exp Epidemiology/ or exp Allied Health Personnel/ or exp Nutrition/ or exp Diets/ or exp Neurodevelopmental Disorders/ or exp Psychotherapeutic Processes/ or exp Counseling/ or exp Counseling Psychology/ or exp Counselors/ or exp Client Centered Therapy/ or exp Management/) and exp Organizations/
22	21 use psyh
23	((care or healthcare or nursing or nurse*) adj3 (organi* or deliv* or service* or model* or communit* or home* or domicil*)).ti,ab.
24	((home* or domicil* or communit* or outreach or access* or special*) adj3 (care or healthcare or center* or centre* or clinic* or service*)).ti,ab.
25	((therapeut* or treatment* or program* or service*) adj5 (need* or individual* or tailor* or care or personal* or anticipatory or enablement or re?enablement or continuity)).ti,ab.
26	((multidisciplin* adj2 team*) or MDT).ti,ab.
27	(treatment* or therapy* or program* or change* or public understanding or intervention* or holistic or rehab* or policy or policies or empower* or disempower* or improve* or standard* or service* or planning or support* or manag* or mainstream* or assist* or provi* or training or funding or network* or quality or local* or communit* or hospital* or home or measure* or scale* or score* or teamwork or aftercare or follow-up* or follow up* or initiative* or surveill* or self-care or life style or ageing or age* or daily or body maintenance or self-care or health*).ti,ab.
28	(epidemiologist* or occupational therapist* or counselor* or nutritionist* or dietician* or orthop?edic* or neurosurgeon* or neurologist* or social worker* or general practitioner or GP or specialist* or surgeon* or physiotherapist* or psychiat* or speech therapist* or language therapist*).ti,ab.
29	(Local Authorit* or Health Board* or health service* or Community Disability Team*).ti,ab.
30	(Canadian Occupational Performance Measure or COPM).ti,ab.
31	or/23-30
32	18 or 20 or 22 or 31
33	16 and 32
34	conference abstract.pt. use oemezd
35	letter.pt. or LETTER/ use oemezd
36	Letter/ use prmz
37	EDITORIAL/ use prmz
38	editorial.pt. use oemezd
39	NEWS/ use prmz
40	exp HISTORICAL ARTICLE/ use prmz
41	note.pt. use oemezd

#	Searches
42	ANECDOTES AS TOPIC/ use prmz
43	COMMENT/ use prmz
44	CASE REPORT/ use prmz
45	CASE REPORT/ use oemezd
46	CASE STUDY/ use oemezd
47	(letter or comment* or abstracts).ti.
48	or/34-47
49	RANDOMIZED CONTROLLED TRIAL/ use prmz
50	RANDOMIZED CONTROLLED TRIAL/ use oemezd
51	random*.ti,ab.
52	or/49-51
53	48 not 52
54	ANIMALS/ not HUMANS/ use prmz
55	ANIMAL/ not HUMAN/ use oemezd
56	exp ANIMALS, LABORATORY/ use prmz
57	exp ANIMAL EXPERIMENTATION/ use prmz
58	exp MODELS, ANIMAL/ use prmz
59	exp RODENTIA/ use prmz
60	NONHUMAN/ use oemezd
61	exp ANIMAL EXPERIMENT/ use oemezd
62	exp EXPERIMENTAL ANIMAL/ use oemezd
63	ANIMAL MODEL/ use oemezd
64	exp RODENT/ use oemezd
65	(rat or rats or mouse or mice).ti.
66	or/53-65
67	33 not 66
68	limit 67 to yr="1990 -Current"

Database: Cochrane Library

Table 9: Last searched on 22 March 2018

ID	Search
#1	MeSH descriptor: [Cerebral Palsy] explode all trees
#2	((cerebral or brain or central) N2 (pal* or paralys?s or pare?s))
#3	((hemipleg* or dipleg* or tripleg* or quadripleg* or unilateral*) N5 spastic*)
#4	((hemipleg* or dipleg* or tripleg* or quadripleg* or unilateral*) N3 ataxi*)
#5	#1 or #2 or #3 or #4
#6	MeSH descriptor: [Delivery of Health Care] explode all trees
#7	MeSH descriptor: [Health Services Administration] explode all trees
#8	MeSH descriptor: [Models, Organizational] explode all trees
#9	MeSH descriptor: [Models, Nursing] explode all trees
#10	MeSH descriptor: [Home Health Nursing] explode all trees

ID	Search
#11	MeSH descriptor: [Community Health Services] explode all trees
#12	MeSH descriptor: [Community Health Nursing] explode all trees
#13	MeSH descriptor: [Health Services Accessibility] explode all trees
#14	MeSH descriptor: [Health Education] explode all trees
#15	MeSH descriptor: [Health Care Rationing] explode all trees
#16	MeSH descriptor: [Health Personnel] explode all trees
#17	MeSH descriptor: [Attitude of Health Personnel] explode all trees
#18	MeSH descriptor: [Patient Care Team] explode all trees
#19	MeSH descriptor: [Health Status Indicators] explode all trees
#20	MeSH descriptor: [Health Status] explode all trees
#21	MeSH descriptor: [Rehabilitation Centers] explode all trees
#22	MeSH descriptor: [Physical and Rehabilitation Medicine] explode all trees
#23	MeSH descriptor: [Neurological Rehabilitation] explode all trees
#24	MeSH descriptor: [Rehabilitation Nursing] explode all trees
#25	MeSH descriptor: [Rehabilitation, Vocational] explode all trees
#26	MeSH descriptor: [Rehabilitation] explode all trees
#27	MeSH descriptor: [Psychiatric Rehabilitation] explode all trees
#28	MeSH descriptor: [Continuity of Patient Care] explode all trees
#29	MeSH descriptor: [Professional-Family Relations] explode all trees
#30	MeSH descriptor: [Primary Health Care] explode all trees
#31	MeSH descriptor: [Patient Acceptance of Health Care] explode all trees
#32	MeSH descriptor: [Quality of Health Care] explode all trees
#33	MeSH descriptor: [Quality of Life] explode all trees
#34	MeSH descriptor: [Mental Health Services] explode all trees
#35	MeSH descriptor: [Health Services Needs and Demand] explode all trees
#36	MeSH descriptor: [Needs Assessment] explode all trees
#37	MeSH descriptor: [Community Mental Health Services] explode all trees
#38	MeSH descriptor: [Public Policy] explode all trees
#39	MeSH descriptor: [Health Policy] explode all trees
#40	MeSH descriptor: [Standard of Care] explode all trees
#41	MeSH descriptor: [Professional Practice] explode all trees
#42	MeSH descriptor: [Medical Audit] explode all trees
#43	MeSH descriptor: [Clinical Audit] explode all trees
#44	MeSH descriptor: [Utilization Review] explode all trees
#45	MeSH descriptor: [State Medicine] explode all trees
#46	MeSH descriptor: [Social Support] explode all trees
#47	MeSH descriptor: [Social Work] explode all trees
#48	MeSH descriptor: [Health Services for the Aged] explode all trees
#49	MeSH descriptor: [Education, Special] explode all trees
#50	MeSH descriptor: [Employment] explode all trees
#51	MeSH descriptor: [Financing, Government] explode all trees

ID	Search
#52	MeSH descriptor: [National Health Programs] explode all trees
#53	MeSH descriptor: [Referral and Consultation] explode all trees
#54	MeSH descriptor: [Hospitalization] explode all trees
#55	MeSH descriptor: [Disability Evaluation] explode all trees
#56	MeSH descriptor: [Disease Management] explode all trees
#57	MeSH descriptor: [Severity of Illness Index] explode all trees
#58	MeSH descriptor: [Transportation of Patients] explode all trees
#59	MeSH descriptor: [Patient Preference] explode all trees
#60	MeSH descriptor: [Outcome and Process Assessment (Health Care)] explode all trees
#61	MeSH descriptor: [Sickness Impact Profile] explode all trees
#62	MeSH descriptor: [Activities of Daily Living] explode all trees
#63	MeSH descriptor: [Self Care] explode all trees
#64	MeSH descriptor: [Exercise Therapy] explode all trees
#65	MeSH descriptor: [Home Care Services] explode all trees
#66	MeSH descriptor: [Complementary Therapies] explode all trees
#67	MeSH descriptor: [Recreation] explode all trees
#68	MeSH descriptor: [Leisure Activities] explode all trees
#69	MeSH descriptor: [Age Factors] explode all trees
#70	MeSH descriptor: [Life Style] explode all trees
#71	MeSH descriptor: [Transition to Adult Care] explode all trees
#72	MeSH descriptor: [Socioeconomic Factors] explode all trees
#73	MeSH descriptor: [Aftercare] explode all trees
#74	MeSH descriptor: [Speech Therapy] explode all trees
#75	MeSH descriptor: [Language Therapy] explode all trees
#76	MeSH descriptor: [Occupational Therapy] explode all trees
#77	MeSH descriptor: [Physical Therapy Modalities] explode all trees
#78	MeSH descriptor: [Behavior Therapy] explode all trees
#79	MeSH descriptor: [Psychotherapy] explode all trees
#80	MeSH descriptor: [Cognitive Therapy] explode all trees
#81	MeSH descriptor: [Orthopedics] explode all trees
#82	MeSH descriptor: [Neurologists] explode all trees
#83	MeSH descriptor: [Counseling] explode all trees
#84	MeSH descriptor: [Counselors] explode all trees
#85	MeSH descriptor: [Social Workers] explode all trees
#86	MeSH descriptor: [Psychiatry] explode all trees
#87	MeSH descriptor: [Dietetics] explode all trees
#88	MeSH descriptor: [Nutritionists] explode all trees
#89	MeSH descriptor: [Neurodevelopmental Disorders] explode all trees and with qualifier(s): [Rehabilitation - RH]
#90	(care or healthcare or nursing or nurse* or center* or centre* or clinic) near (organi* or deliv* or service* or model* or communit* or home* or domicil*)

ID	Search
#91	(therapeut* or treatment* or program* or service*) near (need* or individual* or tailor* or care or personal* or anticipatory or enablement or re?enablement or continuity)
#92	multidisciplin* near team* or MDT
#93	treatment* or therapy* or program* or change* or public understanding or intervention* or holistic or rehab* or policy or policies or empower* or disempower* or improve* or standard* or service* or planning or support* or manag* or mainstream* or assist* or provi* or training or funding or network* or quality or local* or communit* or hospital* or home or measure* or scale* or score* or teamwork or aftercare or follow-up* or follow up* or initiative* or surveill* or self-care or life style or ageing or age* or daily or body maintenance or self-care or health*
#94	epidemiologist* or occupational therapist* or counselor* or nutritionist* or dietician* or orthop?edic* or neurosurgeon* or neurologist* or social worker* or general practitioner or GP or specialist* or surgeon* or physiotherapist* or psychiat* or speech therapist* or language therapist*
#95	{or #6-#94}
#96	#5 and #95 Publication Year from 1990 to 2017

Database: Web of Science

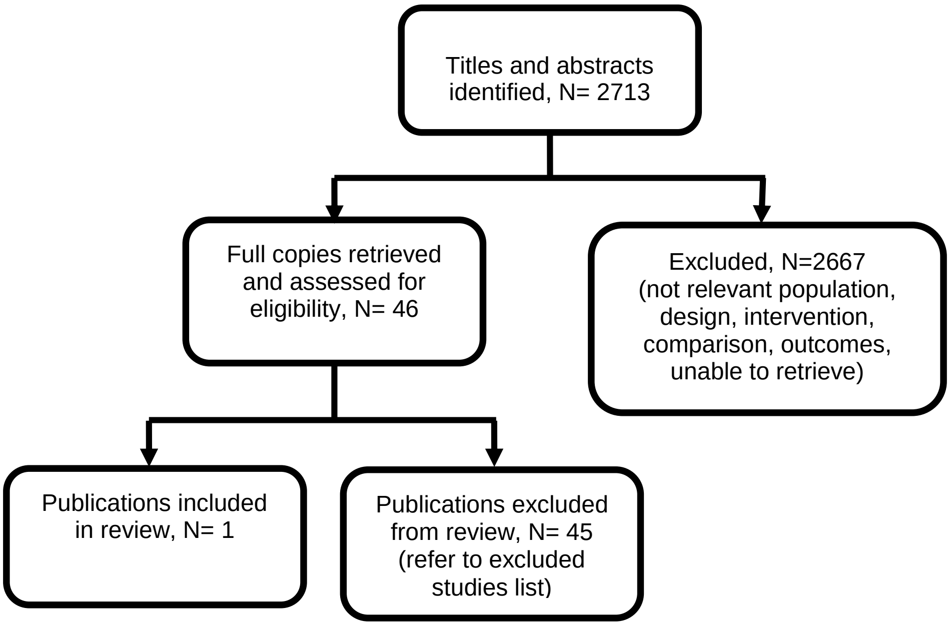
Table 10: Last searched on 22 March 2018

#4	(#3) AND LANGUAGE: (English)
#3	#2 AND #1
#2	ts=healthcare or ts=service* or ts=model* or ts=therapeut* or ts=treatment* or ts=program* or ts=need* or ts=multidisciplin* team* or ts=MDT or ts=change* or ts=public understanding or ts=intervention* or ts=holistic or ts=rehab* or ts=policy or ts=policies or ts=empower* or ts=disempower* or ts=improve* or ts=standard* or ts=planning or ts=support* or ts=manag* or ts=mainstream* or ts=assist* or ts=provi* or ts=training or ts=funding or ts=network* or ts=quality or ts=local* or ts=communit* or ts=hospital* or ts=home or ts=measure* or ts=scale* or ts=score* or ts=teamwork or ts=aftercare or ts=follow-up or ts=follow up or ts=initiative* or ts=surveill* or ts=self-care or ts=life style or ts=ageing or ts=body maintenance or ts=epidemiologist* or ts=occupational therapist* or ts=counsel* or ts=nutritionist* or ts=dietician* or ts=orthop?edic* or ts=neurosurgeon* or ts=neurologist* or ts=social worker* or ts=general practitioner or ts=GP or ts=specialist* or ts=surgeon* or ts=physiotherapist* or ts=psychiat* or ts=speech therapist* or ts=language therapist*
#1	ts=cerebral palsy

Appendix C – Clinical evidence study selection

Clinical evidence study selection for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

Figure 4: Flow diagram of clinical article selection for configuration of services review



Appendix D – Clinical evidence tables

Clinical evidence tables for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

Table 11: Studies included in the evidence review for configuration of services

Study details	Participants	Interventions	Methods	Outcomes and Results	Comments
Full citation Young, Nancy L., Steele, Catherine, Fehlings, Darcy, Jutai, Jeffery, Olmsted, Nancy, Williams, J., Use of health care among adults with chronic and complex physical disabilities of childhood, Disability and Rehabilitation: An International, Multidisciplinary Journal, 27, 1455-1460, 2005 **Ref Id** 657209 **Country/ies where the study was carried out** Canada **Study type**	**Sample size** 345 **Characteristics** Age: mean 21.9 years (range 19 to 26.9) Diagnosis: 58% had CP, 25% SB and 17% ABI GMFCS level: for those with CP 42% had GMFCS I to III and 58% GMFCS > III Learning disability: not reported Communication ability: not reported **Inclusion criteria**	**Interventions** Outcomes were reported according to whether the person had a single primary care doctor or multiple different doctors. The authors thought that the presence of a primary care doctor was an indicator of an adequate level health maintenance and promotion.	**Details** Health records of the included adults were checked for demographics and details of their condition. Outcomes recorded were: frequency of outpatient doctor visits, emergency department visits, the number of in-patient episodes of care and whether or not the person had a primary care doctor. These outcomes were ascertained by linking the medical centre's records to the data from the Ontario Health Insurance Plan and data from the Canadian Institute for Health Information.	**Results** See forest plots	**Limitations** Selection 1) Representativeness of the exposed cohort somewhat representative - only 58% had CP; Canadian definition of primary care doctor may differ to the UK 2) Selection of the non exposed cohort drawn from the same community as the exposed cohort; again service models may differ to the UK 3) Ascertainment of exposure secure record (eg surgical records)

Study details	Participants	Interventions	Methods	Outcomes and Results	Comments
Retrospective cohort study **Aim of the study** To describe health service use among adults with cerebral palsy (CP), spina bifida (SB) or acquired brain injuries in childhood (ABI). **Study dates** 1996 to 1999 **Source of funding** Bloorview Children's Hospital Foundation grant.	Young adults (aged 19 to 27 in 1996) with CP, SB or ABI who had graduated from a single children's treatment centre in Ontario clients. Only those who had received medical care at the centre were included. **Exclusion criteria** Not reported				4) Demonstration that outcome of interest was not present at start of study not applicable Comparability 1) Comparability of cohorts on the basis of the design or analysis study does not controls for any additional factors that might be associated with the outcome Outcome 1) Assessment of outcome record linkage 2) Was follow-up long enough for outcomes to occur yes 3) Adequacy of follow up of cohorts complete follow up - all subjects accounted for Overall quality: low

Study details	Participants	Interventions	Methods	Outcomes and Results	Comments
					Other information

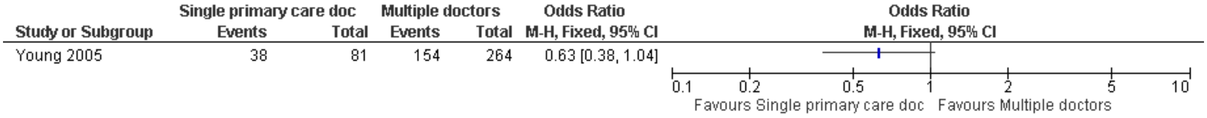
ABI: acquired brain injury; CP: cerebral palsy; GMFCS: general motor function classification system; SB: spina bifida

Appendix K – Forest plots

Forest plots for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

Single primary healthcare provider versus other service configuration

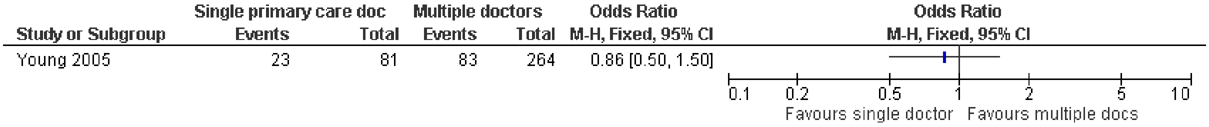
Figure 5: Visit to emergency department within the last 4 years



	Single primary care doc		Multiple doctors		Odds Ratio
Study or Subgroup	Events	Total	Events	Total	M-H, Fixed, 95% CI
Young 2005	38	81	154	264	0.63 [0.38, 1.04]

CI: confidence interval; M-H: Mantel-Haenszel

Figure 6: Inpatient admission within the last 4 years



	Single primary care doc		Multiple doctors		Odds Ratio
Study or Subgroup	Events	Total	Events	Total	M-H, Fixed, 95% CI
Young 2005	23	81	83	264	0.86 [0.50, 1.50]

CI: confidence interval; M-H: Mantel-Haenszel

Appendix F – GRADE tables

GRADE tables for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

Table 12: Clinical evidence profile: Comparison 1: Primary healthcare providers versus other service configuration

Quality assessment							**No of patients**		**Effect**		**Quality**	**Importance**
No of studies	**Design**	**Risk of bias**	**Inconsistency**	**Indirectness**	**Imprecision**	**Other considerations**	**Primary Care doctor**	**Multiple different doctors**	**Relative (95% CI)**	**Absolute**		
HRQOL - not reported												
-	-	-	-	-	-	-	-	-	-	-	-	CRITICAL
Time to treatment - not reported												
-	-	-	-	-	-	-	-	-	-	-	-	CRITICAL
Visit to emergency dept. in past 4 years												
1	observational studies	serious[1]	no serious inconsistency	serious[2,3]	serious[4]	none	38/81 (46.9%)	154/264 (58.3%)	OR 0.63 (0.38 to 1.04)	115 fewer per 1000 (from 236 fewer to 10 more)	VERY LOW	CRITICAL
In-patient admission in the last 4 years (unclear if planned or not)												
1	observational studies	serious[1]	no serious inconsistency	serious[2,3]	serious[4]	none	23/81 (28.4%)	83/264 (31.4%)	OR 0.86 (0.5 to 1.5)	32 fewer per 1000 (from 128 fewer to 93 more)	VERY LOW	CRITICAL
Adverse effects - not reported												
-	-	-	-	-	-	-	-	-	-	-		IMPORTANT
Unplanned residential care admissions - not reported												
-	-	-	-	-	-	-	-	-	-	-		IMPORTANT
Length of hospital stay - not reported												
-	-	-	-	-	-	-	-	-	-	-		IMPORTANT
Mortality - not reported												
-	-	-	-	-	-	-	-	-	-	-	-	IMPORTANT
Satisfaction - not reported												
-	-	-	-	-	-	-	-	-	-	-	-	IMPORTANT

CI, confidence interval; HRQOL, health related quality of life; OR, odds ratio

1 Baseline differences in characteristics or severity of condition not controlled for in analysis
2 Only 58% of sample had cerebral palsy. Unclear whether the Canadian primary care model is applicable to the UK.
3 Unclear whether inpatient episodes were planned or unplanned.
4 Downgraded for imprecision: number of events < 300

Appendix G – Economic evidence study selection

Economic evidence study selection for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

No economic evidence was identified for this review.

Appendix H – Economic evidence tables

Economic evidence tables for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

No economic evidence was identified for this review.

Appendix I – Health economic evidence profiles

Health economic evidence profiles for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

No economic evidence was identified for this review.

Appendix J – Health economic analysis

Health economic analysis for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

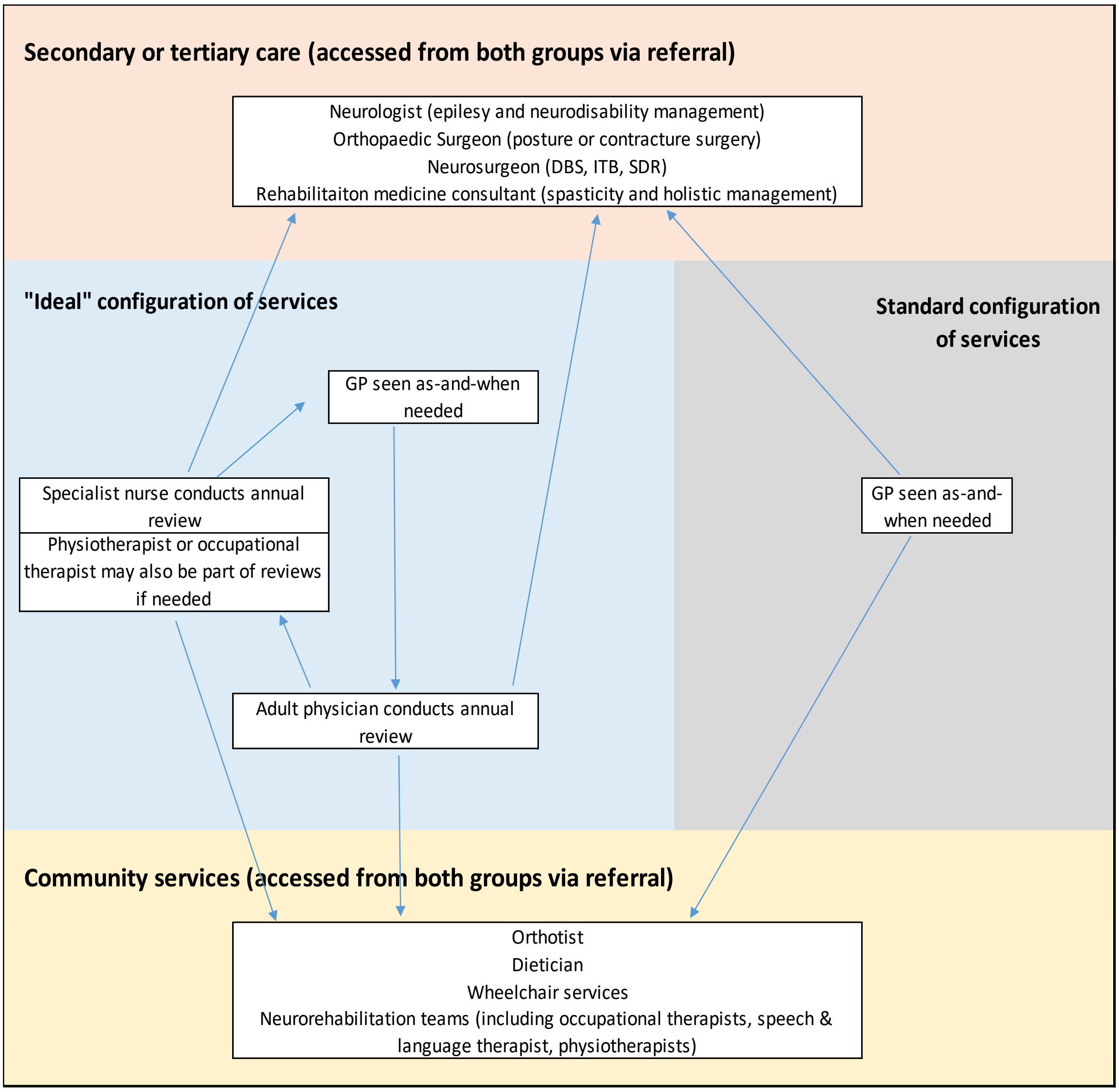
Background

Services for adults with cerebral palsy are limited in the UK. GPs are often the first point of contact, but this can lead to emergency department visits when complications arise. This question has been prioritised by the committee for health economic analysis as recommendations may identify a new configuration of services, or recommend additional routine reviews, potentially affecting all adults with cerebral palsy (approximately 110,000 children and adults in the UK today, according to the Neurological Alliance http://www.neural.org.uk/store/assets/files/20/original/NeuroNumbers.pdf [last accessed 29/05/2018]). As a result, recommendations could lead to a “significant” increase in resource use. The economic search did not identify economic evaluations assessing the cost effectiveness of services and this area is not addressed by any previous NICE guidance.

A conceptual model was developed with the committee to provide a simplified, diagrammatical representation of the care/service pathway that describes the resources, processes and interactions in the delivery of healthcare interventions. This is provided in Figure 7 and described in Table 13.

This exercise established the breadth and complexity of services for adults with cerebral palsy, namely due to the heterogeneity of need which cannot be captured fully in an economic model. For example, there are adults with cerebral palsy who have few, if any medical needs where it may not be cost effective to provide routine reviews. Others however, may experience delays to appropriate management in the absence of routine reviews. Furthermore, a responsive service may be needed by adults with cerebral palsy when unexpected changes occur. As a result, mandatory routine reviews should not be recommended for all adults with cerebral palsy without justification.

Figure 7: Structure of conceptual model



CP – cerebral palsy; DBS – deep brain stimulation; GP – general practitioner; ITB – intrathecal baclofen; SDR – selective dorsal rhizotomy

Table 13: Summary of conceptual model

Service pathway for “ideal “ services in the model (based on current provision for some adults with severe cerebral palsy)
Adults with cerebral palsy will be offered annual review by the GP or practice nurse.
Adults will also see each of an adult physician, specialist nurse and physiotherapist or occupational therapist (3 appointments in total) for a routine review every year, or more frequently if there are active problems. A member of the neurorehabilitation team such as a therapist will also be involved. This is currently not standard practice for all adults with cerebral palsy.

According to the committee those routine reviews would take approximately 30 minutes for each of the three healthcare professionals to conduct (1 hour and 30 minutes total). Additional administration time will also be needed.
Adults also see their GP as-and-when required, but no formal routine reviews are conducted.
Adults with cerebral palsy have access to community, secondary or tertiary care services via a referral.
If regular reviews are recommended for all adults with cerebral palsy, additional training and administration costs will be incurred.
Service pathway for standard configuration of services
Adults see their GP and other health care professionals as-and-when required, but no formal routine reviews are conducted.
Adults with cerebral palsy have access to community, secondary or tertiary care services via a referral.

GP – general practitioner

Model structure

A decision analytic model was developed in Microsoft Excel® (2013) from the perspective of the UK NHS and using 2015/16 costs. The model takes a 1-year time horizon due to the lack of clinical evidence available to extrapolate. Mortality is not considered.

Two outcomes (unplanned hospital visits [emergency department visits] and length of stay in hospital) specified in the protocol were explored in the model. 3 scenarios were considered to explore the differences needed for an “ideal” configuration of services to be cost effective:

1. Differences in emergency department visits excluding subsequent admissions to hospital
2. Differences in length of hospital stay excluding emergency department visits
3. Differences in emergency department visits followed by hospital admissions.

Those scenarios were compared between an “ideal” configuration of services (based on constrained resources) and the standard configuration of services the conceptual model contextualised. The scenarios are presented in detail in Figure 7 and services provided described in Table 13. A summary of how “ideal” services have been simplified are presented in Table 14. Standard service consists solely of individuals visiting their GP when and if needed and also forms part of the configurations of “ideal” services. As this will be for a range of conditions both related and unrelated to cerebral palsy and not expected to differ between the groups it was not included in the model. Both groups would also be able to access secondary, tertiary and community care services either by referral from their GP or by other health care practitioner. Again in the absence of evidence to the contrary the use of these resources was considered identical between the groups and again was not included in the economic model.

Table 14: Configuration of ideal services in the economic model

Health care Professional	Length of Visit
Adult physician	30 minute appointment annually
Specialist Nurse	30 minute appointment annually
Physiotherapist/Occupational Therapist	30 minute appointment annually

Clinical effectiveness

No relevant comparative clinical data was identified in the clinical evidence review. Therefore threshold and "what-if" analyses were undertaken to identify the mean differences required for "ideal" service to be considered cost effective compared to standard configuration of services. Relative differences such as relative risks are not reported as baseline data could not be applied in the absence of relevant UK evidence.

Some studies identified in the clinical evidence review compared the outcomes for children and adults. If the outcomes produced by children's services could be considered as a proxy for "ideal" services, and adult services for standard services, an ICER could be calculated. However, it was evident that the outcomes from those studies would be confounded by factors such as age. As a result, the committee agreed it would be inappropriate to make such a comparison.

Health-related quality of life

The QALY is NICE's preferred measure of benefit for economic evaluation. This is because it can be seen as a generic measure of health which allows a comparison across treatments which affect different dimensions of health.

The QALY reflects the 2 principle objectives of health care:

- increased longevity;
- increased quality of life.

Estimating a QALY involves placing a quality of life weight on a particular health state. This quality weight, or "utility" lies between 0 and 1, where 1 denotes full or 'perfect health' and 0 denotes death. Based on a need for consistency across technology appraisals and guidelines, NICE favours a generic, preference based measure reported by patients to value health states such as the EQ-5D which comes with pre-existing utility values obtained from a representative sample of the UK general population. However, other measures and value sets are available.

The utility value is multiplied by the length of life measured in years to produce a QALY, such that 1 year spent in full health is 1 QALY. Similarly, the disutility can be multiplied by the length of life measured in years to produce a QALY loss.

A baseline utility was not applied in this model as adults with cerebral palsy have a wide range of clinical presentations and functional limitations, resulting in a range of possible utilities. To overcome this, the (incremental) difference in QALY loss was estimated. The model did not account for any potential floor or ceiling effects from people who had either a near perfect or very low quality of life at baseline and therefore could not experience as large increases/decreases in utility. Given the impact upon coordination and movement of cerebral palsy it was considered unlikely that any of the cohort would have a utility value at baseline near to 1. Whilst those with the very severest form of cerebral palsy would have a low utility score, given severe difficulties with movement, breathing and pain as well as other dimensions, this group would already be under regular review of their health needs most likely greater than annually. It was therefore considered that the majority of the cohort would have a baseline where the QALY detriments described below would not pass the extremes at either end of the scale.

QALY loss from emergency department visits and hospital stays

A search was undertaken in the CEA Tufts registry to identify utility values related to an emergency department visit and hospital stay.

A utility value of 0.513 was identified from Dixon 2009 who elicited preferences from people in the UK using the EQ-5D 28 days following an emergency call.

Patients aged 55 to 64 in Dixon 2009 would have a baseline utility of 0.80 according to the population norms reported by Dolan 1997. Taking this baseline into account, patients would have a disutility of 0.287 (0.80 minus 0.513) when they visit the emergency department.

Assuming an emergency department visit lasts 1 day, the QALY loss would equal 0.0008 (0.287* (1/365)).

Adults with cerebral palsy may be admitted to hospital for a variety of reasons with concomitant differences in their treatment. To account for this, disutilities associated with the 4 most common reasons adults with cerebral palsy are admitted (Young 2011) were used to inform the model (Table 15).

Sullivan 2011 derived a catalogue of CCC and ICD-9 scores for the EQ-5D using censored least absolute deviation (CLAD) regressions. They used data from 2000, 2001, 2002, and 2003 Medical Expenditure Panel Survey (MEPS) which contained 79,522 individuals with EQ-5D indexes that were estimated using the scoring function based on UK community preferences (Dolan 1997). Each condition coefficient from the regression analyses represented the marginal disutility associated with that condition. Those coefficients also represented the marginal decrement in EQ-5D index scores for each condition after controlling for age, comorbidity, gender, race, ethnicity, income, and education.

Sullivan 2011 was chosen to inform the model to promote consistency, as other studies that reported conditions separately in the CEA Tufts Registry had disparate populations, making it difficult to compare the impact across the studies and conditions. Moreover, the scores in Sullivan 2011 were applicable to a UK population, using NICE’s preferred method of elicitation (EQ-5D).

The length of a hospital stay is variable and the QALY loss (disutility x duration) will increase with the length of stay (LOS). A patient’s quality of life could increase towards the end of their stay if they became well, but may not if a longer duration was associated with complications. In the model, a linear relationship was assumed (Figure 8) as no evidence was identified to suggest that other relationships were plausible. The QALY loss is calculated using this formula in the model: -0.0563*(LOS/365).

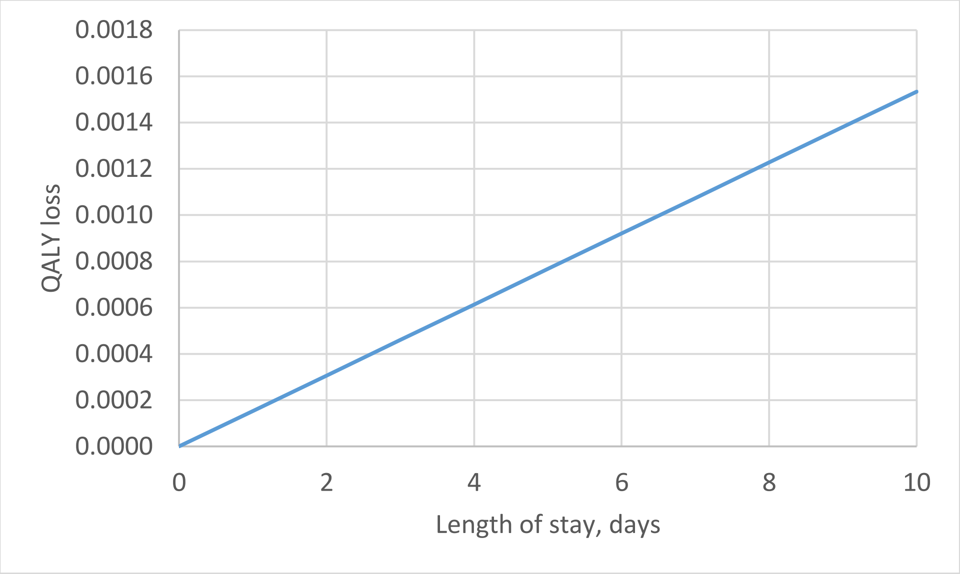
Table 15: Quality adjusted life year (QALY) loss

Reason for admission	Disutility	Weight	Proportion of admissions in Young 2011
ICD-9 519 Other Respiratory System Diseases	-0.0372	35%	15.5% related to pneumonia (no ICD code related to pneumonia reported by Sullivan, “Other Respiratory System Diseases” used as a proxy)
ICD-9 345 Epilepsy	-0.0399	17%	7.5%
ICD-9 V40 Mental/Behavioural Problems	-0.0946	15%	6.4%

Reason for admission	Disutility	Weight	Proportion of admissions in Young 2011
ICD-9 537 Other Gastrointestinal Disorders	-0.0684	33%	14.4% (includes lower GI 6.4%, upper GI 3.7% and other GI 4.3%)
Weighted disutility by reason	**-0.0563**	**-**	**-**

GI: gastrointestinal; ICD-9: international classification of diseases (9th edition)

Figure 8: Illustration of relationship between QALY loss for a hospital stay and length of stay



QUALY – quality adjusted life years

Resource and cost use

Staffing

Curtis (PSSRU, Personal Social Services Research Unit) 2016 considers the following costs when calculating the cost per hour of healthcare professionals:

- Wages
- Salary on-costs (employer's national insurance plus contribution to superannuation)
- Staff overheads (administration and estates staff)
- Non-staff overheads (costs to the provider for office, publishing, training courses and conferences, supplies and services for clinical and general use, and utilities such as water, gas and electricity)
- Capital overheads (based on the new-build and land requirements of NHS hospital facilities)

Those costs from Curtis 2016 for healthcare professionals that care for adults with cerebral palsy are provided in Table 16, per working hour.

Table 16: healthcare professionals providing care for adults with cerebral palsy

Health care professional	Cost
“Core” providing regular reviews	
Specialist nurse (band 6)	£44
Physiotherapist (band 6)	£45
Adult physician (consultant medical)	£135
“Extended” available as-and when-needed	
GP cost per hour of patient contact	£236
GP per surgery consultation 9.22 lasting minutes	£36
Consultant medical (neurologist, rehabilitation medicine or physiatrist)	£135
Consultant surgical (orthopaedic surgeon or neurosurgeon)	£137
Dietician (band 6)	£44
Speech and language therapist (band 6)	£44
Occupational therapist (band 6)	£45
Orthotist (band 6)	£45
Social worker	£79
Wheelchair services (NHS Reference Costs 2015/16, WC11 cost per review of all needs)	£152

NHS: National health service

The healthcare professionals conducting routine reviews in the model include a specialist nurse, adult physician and physiotherapist (or occupational therapist). Reviews with a GP are not considered in the model as they are performed as-and-when required under both configuration of services. The committee agreed this was in line with some services currently in place for adults with cerebral palsy. For example, Field 2010 describe how the Westmead Hospital Adult Physical Disability Clinic (WHPDC) is staffed by a rehabilitation physician, physiotherapist and occupational therapist.

According to the committee annual reviews would take approximately 30 minutes per healthcare professional to conduct, with up to an additional 30 minutes of administration following the review. Based on this, the annual cost to provide routine reviews with an adult physician, specialist nurse and physiotherapist (or occupational therapist) is approximately £224 per review per adult with cerebral palsy.

The results of all of these eventualities cannot be reported with ease, although the model developed for this review allows the user to select the health care professionals conducting reviews, number of reviews per year and duration of reviews. As a result, the frequency of most interest to the committee (annual reviews) was discussed in greater detail, whilst the results of other scenarios are provided in Table 16 and/or reported narratively in the discussion sections.

If access to specialist reviews is recommended for all adults with cerebral palsy, additional training and administration costs will be incurred as there are not enough adult physicians and physiotherapists with expertise in cerebral palsy to undertake regular reviews across England and Wales. Those implementation costs will be considered as part of NICE’s assessment of the guideline’s resource impact.

Cost of unplanned hospital care

The cost of a visit to the emergency department was taken from NHS Reference Costs 2015/16 (ASS01 ambulance services, see and treat or refer £181 per incident). This code measured the number of incidents, following emergency or urgent calls, resolved with the patient being treated and discharged from ambulance responsibility on scene, or incidents where ambulance trust healthcare professionals on scene refer (but do not convey) the patient to any alternative care pathway or provider.

As stated previously, the reasons for an admission can vary. For consistency with QALY calculations, currency codes for non-elective inpatient care were based on the common reasons adults with cerebral palsy were admitted (Young 2011). To capture the impact of the length of stay in the model, the initial cost of the admission was taken from a short stay, with subsequent days added using “excess bed days”.

Table 17 provides the methods and costs of inpatient care applied in the model, whilst the relationship between the cost and length of stay is illustrated in Table 17.

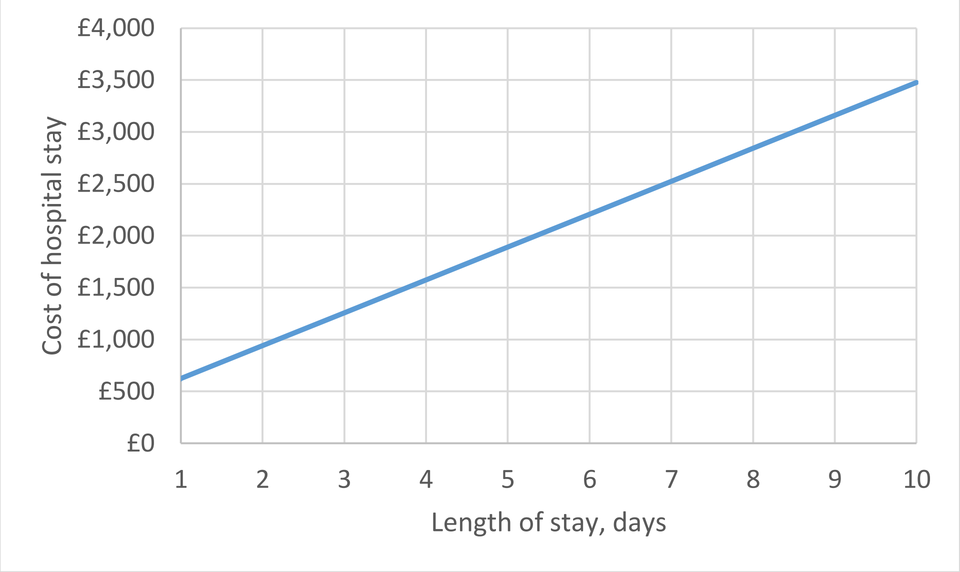
Table 17: Cost of inpatient care

Reason for admission	NHS Reference Costs 2015/16 currency codes	Weight (Table 15)	Non-elective short stay cost applied to the first day of care [a]	Non-elective, excess bed day costs [a]
Pneumonia	DZ11K : DX11V	35%	£504	£259
Epilepsy	AA26C : AA26H	17%	£459	£297
Mental illness	SPHMSOTHAPC	15%	£429	£429
Lower, upper or other GI	FZ2G : FZ24J	33%	£923	£341
Weighted cost by reason	**-**	**-**	**£623**	**£317**

GI: gastrointestinal; NHS: National Health Service

(a)Cost weighted by the Finished Consultant Episodes (FCEs) for each code

Figure 9: Illustration of relationship between the cost of a hospital stay and length of stay (LOS)



Cost effectiveness

If there is strong evidence that an intervention (or service) dominates the alternatives (that is, it is both more effective and less costly), it should normally be recommended. However, if an intervention is more effective but also more costly than another, then the ICER should be considered, to ascertain whether the additional costs represent good value for money for a resource constrained health service.

The cost effectiveness of a healthcare intervention is determined by the opportunity cost of the health foregone on the basis that, with a fixed health care budget, any newly funded intervention would displace the least cost effective treatment currently provided. In the UK, NICE typically uses a threshold of £20,000 per QALY as a benchmark for the opportunity cost of health foregone from the least cost effective treatment currently provided on the NHS.

An ICER below £20,000 per QALY would generally be considered cost effective, whereas an ICER above £30,000 per QALY would generally not be considered cost effective without additional justifications. The committee was asked to consider:

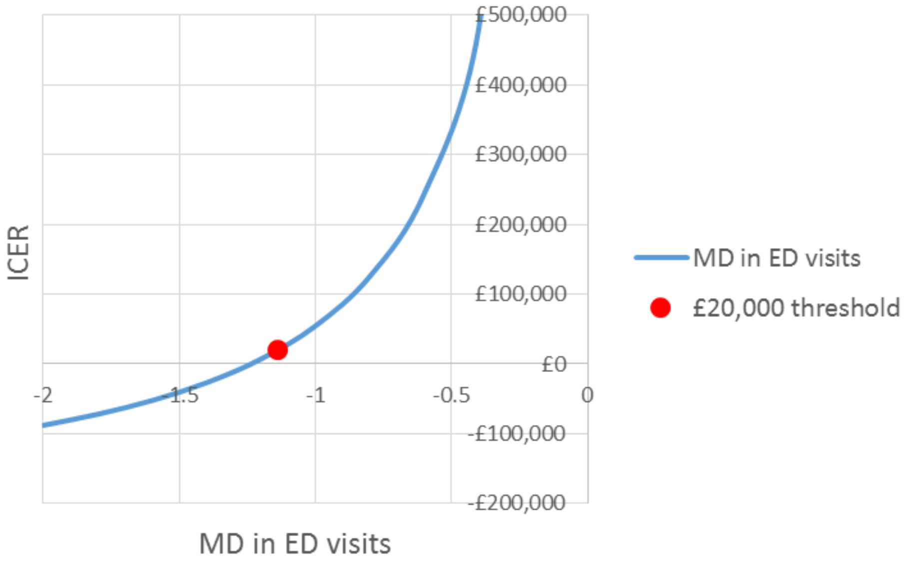
- the degree of certainty around the ICER;
- limitations to the generalisability of the evidence for effectiveness;
- whether the assessment of the change in quality of life has been inadequately captured, and may therefore misrepresent, the health gain or loss;
- whether the intervention is an innovation that adds demonstrable and distinct substantial benefits that may not have been adequately captured in the measurement of health gain.

Results

Emergency department visits excluding subsequent admissions to hospital

When a threshold analysis was conducted on the mean difference in emergency department visits, “ideal” services would be considered cost effective at a threshold of £20,000 per QALY if the number of visits could be reduced by at least 1.14 per year. ICERs are illustrated in Figure 10 with mean differences ranging from 0 to -2 (no difference in visits to 2 less incurred by “ideal” services).

Figure 10: Threshold analysis for ED visits

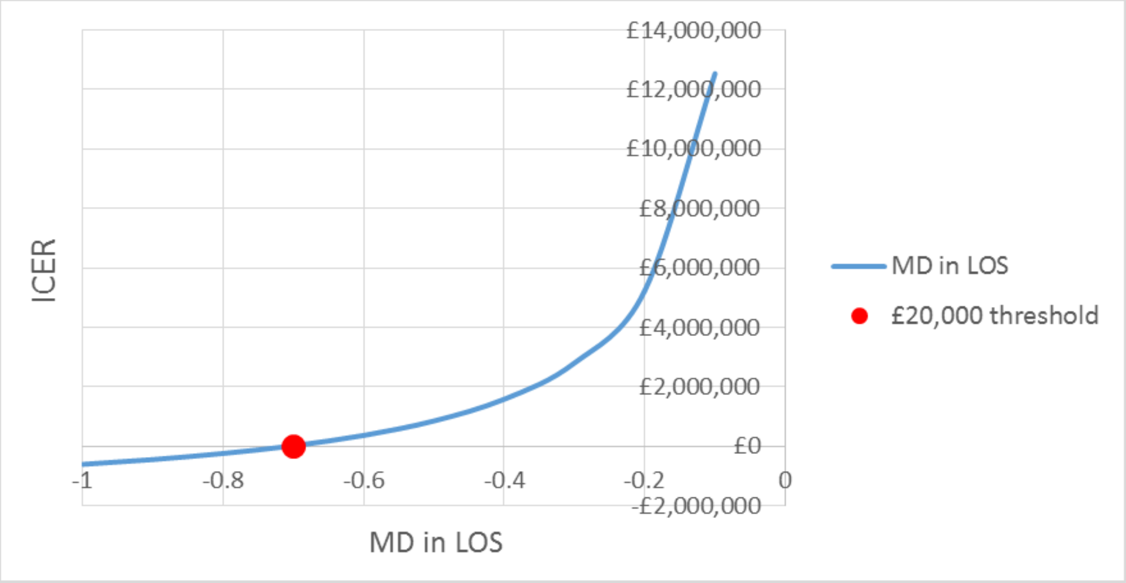


ED: emergency department; ICER: incremental cost effectiveness ratio; MD – mean difference

Admissions to inpatient care excluding emergency department visits

When a threshold analysis was conducted on the mean difference in the length of stay, “ideal” services would be considered cost effective at a threshold of £20,000 per QALY if the number of days could be reduced by at least 0.70. ICERs are illustrated in Figure 11 with mean differences ranging from 0 to -1 (no difference in the length of stay to 1 less day incurred by “ideal” services).

Figure 11: Threshold analysis for LOS



ICER: incremental cost effectiveness ratio; LOS: length of hospital stay; MD: mean difference

Scenario analysis

Alternative scenarios considering the mean difference in emergency department visits or the length of hospital stay needed for "ideal" services to be the preferred option at a threshold of £20,000 per QALY were explored. Those outcomes were considered independently. The results from a dependent analysis – including admissions following a visit to the emergency department are not reported due to the high uncertainty comparing those results and relationships to clinical practice. The list of scenarios in Table 18 is not exhaustive as current practice is variable according to need, geographical location and resources.

Table 18: Results of scenario analyses

Scenario [a]	MD in ED visits	MD in LOS
Base-case: reviews 1x year, 30 minutes in duration+30 minutes administration, conducted by an adult physician, specialist nurse and physiotherapist (or occupational therapist) compared with no routine reviews in standard care	-1.14	-0.70
Reviews as in base case but conducted 3x year	-3.42	-2.10
Reviews as in base case but conducted 0.6x year	-0.68	-0.42
Reviews as in base case but excluding an adult physician	-0.45	-0.28
Reviews 90 minutes (60 minutes+30 minutes administration) in duration	-1.71	-1.05
Standard services include 1 review with a GP lasting 18.44 minutes (lasting 2 standard consultation slots) plus a prescription cost b	-0.63	-0.39

ED: Emergency Department; LOS: length of hospital stay; MD: mean difference
(c) *Holding all other inputs constant with base-case assumptions*
(d) *£28 prescription costs per consultation (PSSRU 2016)*

A geriatrician, who was co-opted to the guideline, working in Edinburgh provided the committee with data that assessed the difference in unplanned hospital attendances, before and after a special needs clinic was implemented. That clinic was staffed by specialist nurses and physiotherapists, who reviewed adults with special needs annually. Before the clinic was implemented, patients had 1.22 emergency department attendances or acute admissions a year. After the clinic was implemented this was reduced by 0.58 to 0.64 per year. Based on the assumptions in the base-case, a difference of 0.58 in emergency department visits alone would not outweigh the cost to provide an “ideal” service. However, if those visits led to an inpatient stay, or did not include an adult physician, for example, “ideal” services may be considered cost effective.

Emergency department visits followed by a hospital stay

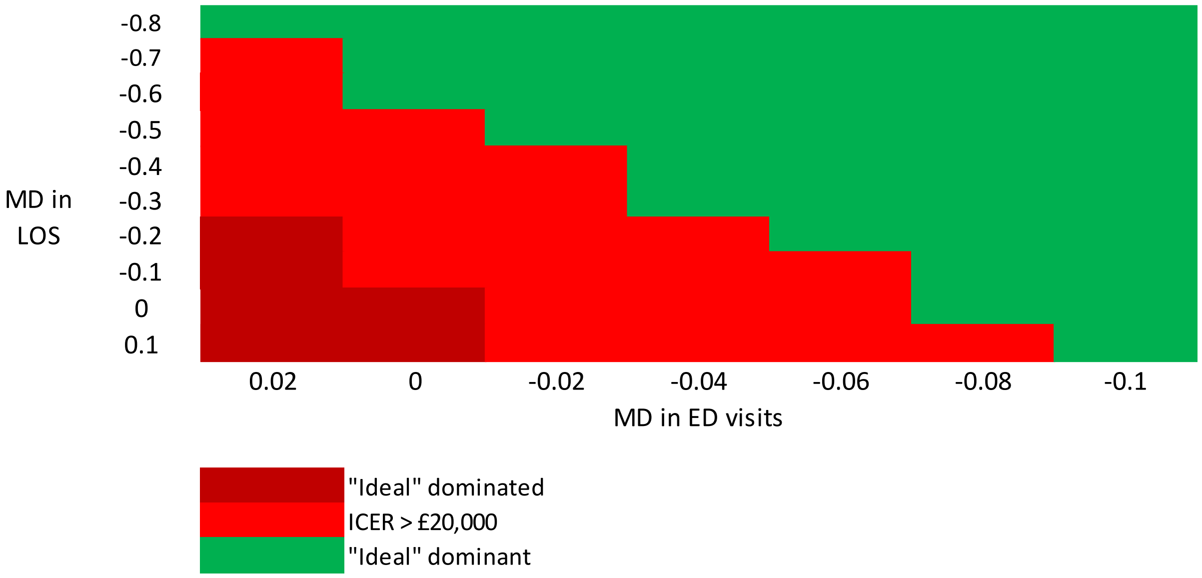
The results of varying the number of emergency department visits and length of hospital stay simultaneously are illustrated in Figure 12, using the base-case scenario. This analysis assumed that 100% of visits to the emergency department lead to an admission.

If we can assume “ideal” services reduced the length of stay by at least 0.7 days, “ideal” services could be considered cost effective compared to standard services, if “ideal” services do not increase the number of emergency department visits compared to standard services.

Figure 12 also shows that if the number of emergency department visits can be reduced by at least 0.08 per year (or, in other words, 1 less visit every 12.5 years), “ideal” services will be cost effective if the length of stay following “ideal” services is not greater than standard services.

The threshold analysis focuses on negative mean differences that favour “ideal” services as they are expected to reduce the number of visits to the emergency department and the length of hospital stays compared to standard services. However, it is important to note that “Ideal” services can be dominated (less effective and more expensive) by standard services when the mean difference in emergency department visits, when the difference in length of stay between two service models is positive.

Figure 12: Threshold analysis varying the mean difference in emergency department visits and length of hospital stay



ED: emergency department; ICER: incremental cost effectiveness ratio; LOS: length of hospital stay; MD: mean difference

Discussion

Using QALYs as the measure of effectiveness incorporates changes in morbidity and mortality and allows broad comparisons across all health care interventions provided by the NHS. Even though no clinical evidence was identified, the threshold type of analyses conducted allowed the committee to consider how much better an “ideal” configuration must be compared to current practice, to be considered a cost effective use of NHS resources.

Assuming 100% of emergency department visits lead to an admission may overestimate the proportion seen in practice. Moreover, the severity of an incident could be reduced by specialist routine reviews in “ideal” practice. However, no evidence was identified to support such a relationship and reasons to visit the emergency department may be independent (unavoidable) of services received prior to the incident.

GP visits were not considered in the base-case comparison as visits to the GP are as-and-when needed for all adults with cerebral palsy. Moreover, no evidence was identified to infer how they might differ between configurations. However, it is important to note that visits to the GP could be more frequent in the absence of specialist routine reviews which could increase the cost effectiveness of “ideal” services if the incremental cost reduces.

Other outcomes in the protocol include residential care admissions and time to appropriate treatment, amongst others. Those outcomes were not included in the model as they have complex and dependent relationships with other outcomes that could not be meaningfully added to the model in the absence of data.

Adults with cerebral palsy differ in their needs which limits the applicability of an analysis that fits the average adult. For completeness, the model allowed the user to define inputs including the configuration of services, but a pragmatic number of comparisons were made and reported. Moreover, the “ideal” configuration of services is subjective and current practice is variable which reiterates the need to produce recommendations for subgroups as one configuration will not be cost effective for all adults at all times.

Conclusions

Overall, no comparative clinical evidence was identified so we cannot say with certainty which configuration of services will be cost effective, or what services or personnel should be included in the configuration. To address this uncertainty, the committee could consider subgroups of adults with cerebral palsy where specialist routine reviews are needed, as the benefits of a review will vary according to the person’s needs which may change with time.

A summary of the committee’s discussion is provided in the rationale and impact section of the guideline.

Appendix K – Excluded studies

Clinical and economic list of excluded studies for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

Clinical studies

Figure 13: Excluded clinical studies for service configuration

Excluded studies – F1 What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?	
Study	**Reason for Exclusion**
Beecham, J., O'Neill, T., Goodman, R., Supporting young adults with hemiplegia: services and costs, Health & Social Care in the Community, 9, 51-9, 2001	Describes health service use and costs of supporting a nationally representative sample of young adults with hemiplegic cerebral palsy. Non-comparative
Berens, J. C., Peacock, C., Implementation of an academic adult primary care clinic for adolescents and young adults with complex, chronic childhood conditions, Journal of Pediatric Rehabilitation Medicine, 8, 3-12, 2015	Describes speciality services used in a transition medicine clinic by those with CP - mean age 20.6 years. No comparison with any other service model.
Bingham, S. C., Beatty, P. W., Rates of access to assistive equipment and medical rehabilitation services among people with disabilities, Disability & Rehabilitation, 25, 487-90, 2003	Compares US health plans: managed care versus fee-for-service in terms of access to assistive equipment and physical rehab when needed. Results combined for cerebral palsy, multiple sclerosis, and spinal cord injury
Bolger, A., Vargus-Adams, J., McMahon, M., Transition of Care in Adolescents With Cerebral Palsy: A Survey of Current Practices, PM and R, 9, 258-264, 2017	Reports a survey of transition-of-care practices in paediatric CP clinics in the USA.
Buzio, A., Morgan, J., Blount, D., The experiences of adults with cerebral palsy during periods of hospitalisation, Australian Journal of Advanced Nursing, 19, 8-14, 2002	Non comparative study: inpatient survey of adults with CP in Australia. Describes experience of care.
Campbell, Fiona, Biggs, Katie, Aldiss, Susie K, O'Neill, Philip M, Clowes, Mark, McDonagh, Janet, While, Alison, Gibson, Faith, Transition of care for adolescents from paediatric services to adult health services, Cochrane Database of Systematic Reviews, 2016	Systematic review of transition from paediatric to adult health care - no CP studies.
Cathels, B. A., Reddihough, D. S., The health care of young adults with cerebral palsy, Medical Journal of Australia, 159, 444-446, 1993	Age 15 to 25. Compares contact with healthcare professionals between school leavers and those still in school.
Ciccarelli, M. R., Brown, M. W., Gladstone, E. B., Woodward, J. F., Swigonski, N. L., Implementation and sustainability of statewide transition support services for youth with intellectual and physical disabilities, Journal of Pediatric Rehabilitation Medicine, 7, 93-104, 2014	Describes implementation of a transition support service for adolescents with disabilities. Non comparative, 31/71 had CP.

Excluded studies – F1 What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

Study	Reason for Exclusion
Colver, A. F., Merrick, H., Deverill, M., Le Couteur, A., Parr, J., Pearce, M. S., Rapley, T., Vale, L., Watson, R., McConachie, H., Study protocol: longitudinal study of the transition of young people with complex health needs from child to adult health services, BMC Public Health, 13, 675, 2013	Study protocol only
Darrah, J., Magil-Evans, J., Adkins, R., How well are we doing? Families of adolescents or young adults with cerebral palsy share their perceptions of service delivery, Disability and Rehabilitation, 24, 542-549, 2002	Qualitative study, does not compare healthcare service models.
Darrah, Johanna, Magill-Evans, Joyce, Galambos, Nancy L., Community services for young adults with motor disabilities-A paradox, Disability and Rehabilitation: An International, Multidisciplinary Journal, 32, 223-229, 2010	Qualitative study, does not compare service models.
de Oliveira Andrade, P. M., de Oliveira Ferreira, F., Haase, V. G., Multidisciplinary perspective for cerebral palsy assessment after an international, classification of functioning, disability and health training, Developmental Neurorehabilitation, 14, 199-207, 2011	Evaluation of training healthcare professionals on the International Classification of Functioning, Disability and Health checklist.
Elrod, C. S., DeJong, G., Determinants of utilization of physical rehabilitation services for persons with chronic and disabling conditions: an exploratory study, Archives of Physical Medicine & Rehabilitation, 89, 114-20, 2008	Factors related to the need for and the receipt of physical rehabilitation services in the USA. Compares healthcare funding source (Medicare, Medicaid, and Private) but not service models.
Field,B., Scheinberg,A., Cruickshank,A., Health care services for adults with cerebral palsy, Australian Family Physician, 39, 165-167, 2010	Non-comparative study. Describes Westmead Hospital Adult Physical Disability Clinic (in NSW) which provides continuity of care for patients discharged from The Children's Hospital at Westmead.
Hagberg,B., Lessons and indications from three decades of West-Swedish Cerebral Palsy data, Neuropediatrics, 31, 284-286, 2000	Retrospective review of a Swedish population based CP study. No comparison of service models.
Hemsley, B., Georgiou, A., Carter, R., Hill, S., Higgins, I., van Vliet, P., Balandin, S., Use of the My Health Record by people with communication disability in Australia: A review to inform the design and direction of future research, Health Information Management Journal, 45, 107-115, 2016	Expert review about Personally Controlled Electronic Health Records in Australians with disabilities.
Hilberink,S.R., Roebroeck,M.E., Nieuwstraten,W., Jalink,L., Verheijden,J.M., Stam,H.J., Health issues in young adults with cerebral palsy: towards a life-span perspective, Journal of Rehabilitation Medicine, 39, 605-611, 2007	Study demonstrates decreased use of allied healthcare services in 25-36 year olds compared to 18 year olds. The configuration of services was not reported.
Himmelmann, K., Sundh, V., Survival with cerebral palsy over five decades in western Sweden,	Does not compare healthcare service models.

Excluded studies – F1 What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?	
Study	**Reason for Exclusion**
Developmental Medicine & Child Neurology, 57, 762-7, 2015	
Hsieh, K., Heller, T., Freels, S., Residential Characteristics, Social Factors, and Mortality Among Adults With Intellectual Disabilities: Transitions Out of Nursing Homes, Intellectual and Developmental Disabilities, 47, 447-465, 2009	45% had CP. Some factors were associated with lower mortality (facility size, environmental diversity, community integration)
Kang, T., Harrington, C., Variation in types of service use and expenditures for individuals with developmental disabilities, Disability and Health Journal, 1, 30-41, 2008	19% had CP, results not reported separately.
Kembhavi,G., Darrah,J., Payne,K., Plesuk,D., Adults with a diagnosis of cerebral palsy: A mapping review of long-term outcomes, Developmental Medicine and Child Neurology, 53, 610-614, 2011	Expert review
Kroll, T., Neri, M. T., Use of primary prevention services among male adults with cerebral palsy, multiple sclerosis, or spinal cord injury in managed care and fee-for-service, Managed Care Quarterly, 12, 6-10, 2004	Compares satisfaction with managed health care vs. fee-for-service in USA. 28% had CP - results not reported separately.
Kroll,T., Beatty,P.W., Bingham,S., Primary care satisfaction among adults with physical disabilities: the role of patient-provider communication, Managed Care Quarterly, 11, 11-19, 2003	Compares use of primary prevention services (e.g. general medical exam, blood pressure check etc.) for managed health care vs. fee-for-service. 23% had CP - results not reported separately.
Li Pi Shan, R. S., Chrusch, W. M., Linassi, A. G., Sankaran, R., Munchinsky, J., Reuse and refurbish: A cost savings delivery model for specialized seating, Archives of Physical Medicine and Rehabilitation, 93, 1286-1288, 2012	Non-comparative description of a specialized seating service model in Canada.
Lifshitz, H., Merrick, J., Morad, M., Health status and ADL functioning of older persons with intellectual disability: community residence versus residential care centers, Research in Developmental Disabilities, 29, 301-15, 2008	Compares morbidity according to type of residence - but not separately for CP
Linroth, R., Meeting the needs of young people and adults with childhood-onset conditions: Gillette Lifetime Specialty Healthcare, Developmental Medicine & Child Neurology, 51 Suppl 4, 174-7, 2009	Non comparative study. Describes Gillette Specialty Healthcare in-patient unit for adults with CP.
McDowell, B., Duffy, C., Parkes, J., Service use and family-centred care in young people with severe cerebral palsy: A population-based, cross-sectional clinical survey, Disability and Rehabilitation: An International, Multidisciplinary Journal, 37, 2324-2329, 2015	Study demonstrates decreased use of healthcare services in 19-27 year olds compared to 12-18 year olds. The configuration of services was not reported.
Middleton, M. J., Kitchen, S. S., Factors affecting the involvement of day centre care staff in the delivery of physiotherapy to adults with intellectual disabilities: An exploratory study in one London borough, Journal of	Reports factors affecting physiotherapy delivery in 4 day centres. Unclear what proportion of service users had CP.

Excluded studies – F1 What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?	
Study	**Reason for Exclusion**
Applied Research in Intellectual Disabilities, 21, 227-235, 2008	
Molics, B., Jaromi, M., Endrei, D., Zemplenyi, A., Boncz, I., Age and Gender Distribution of Outpatient Care Physiotherapy Services for Cerebral Palsy and Other Paralytic Syndromes in Hungary, Value in Health, 17, A810-A810, 2014	Describes use of out-patient physiotherapy services by people with CP in Hungary. Abstract only
Morgan, P. E., Soh, S. E., McGinley, J. L., Health-related quality of life of ambulant adults with cerebral palsy and its association with falls and mobility decline: a preliminary cross sectional study, Health & Quality of Life Outcomes, 12, 132, 2014	Does not compare service models. Reports HRQoL in adults with CP after falls / declining mobility.
Morgan, Prue, Pogrebnoy, Dina, McDonald, Rachael, Health service experiences to address mobility decline in ambulant adults ageing with cerebral palsy, Journal of Intellectual & Developmental Disability, 39, 282-289 8p, 2014	Qualitative study
Morgan, Prue, Williams, Cylie, Tracy, Jane, McDonald, Rachael, Development of a tool to guide clinical decision making in the management of physical function in ambulant adults with cerebral palsy, Journal of Developmental and Physical Disabilities, 28, 785-801, 2016	Describes development of a clinical guideline / algorithm for assessment and management of physical function in adults with CP.
Ng, S. Y., Dinesh, S. K., Tay, S. K., Lee, E. H., Decreased access to health care and social isolation among young adults with cerebral palsy after leaving school, Journal of Orthopaedic Surgery, 11, 80-9, 2003	Study demonstrates decreased use of healthcare services in school leavers (mean age 20) compared with those still in school (mean age 17). The configuration of services was not reported.
Nieuwenhuijsen, C., Van der Laar, Y., Donkervoort, M., Nieuwstraten, W., Roebroeck, M., Stam, H. J., Unmet needs and health care utilization in young adults with cerebral palsy, Disability and Rehabilitation, 30, 1254-1262, 2008	Non comparative study
Palsbo, S. E., Diao, G. Q., The Business Case for Adult Disability Care Coordination, Archives of Physical Medicine and Rehabilitation, 91, 178-183, 2010	Study estimates the costs over time of care coordination for adults with CP in the USA. Non-comparative.
Park, M. S., Kim, S. J., Chung, C. Y., Kwon, D. G., Choi, I. H., Lee, K. M., Prevalence and lifetime healthcare cost of cerebral palsy in South Korea, Health Policy, 100, 234-8, 2011	Does not compare services.
Perez, C. M., Ball, S. L., Wagner, A. P., Clare, I. C. H., Holland, A. J., Redley, M., The incidence of healthcare use, ill health and mortality in adults with intellectual disabilities and mealtime support needs, Journal of Intellectual Disability Research, 59, 638-652, 2015	Non comparative. 30% had CP - results not reported separately.
Prouse, P., Ross-Smith, K., Brill, M., Singh, M., Brennan, P., Frank, A., Community support for young	Study reports healthcare costs in Harrow, N=10 with CP.

Excluded studies – F1 What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?	
Study	**Reason for Exclusion**
physically handicapped people, Health Trends, 23, 105-9, 1991	
Roebroeck, M. E., Van Den Bergemons, H. J. G., Nieuwenhuijsen, C., Hilberink, S. R., Van Der Slot, W. M. A., Van Meeteren, J., Stam, H. J., Innovating transition and lifespan care for people with cerebral palsy, Developmental medicine and child neurology, 52, 74, 2010	Abstract only - survey reports decreased rehab care after leaving childhood CP services.
Termsarasab, P., Medical treatment of dyskinetic cerebral palsy: translation into practice, Developmental Medicine and Child Neurology, 59, 1210-1210, 2017	This is a commentary is on the systematic review by Masson 2017 "Efficacy of oral pharmacological treatments in dyskinetic cerebral palsy: a systematic review"
Wright, Alice E., Robb, James, Shearer, Morven C., Transition from paediatric to adult health services in Scotland for young people with cerebral palsy, Journal of Child Health Care, 20, 205-213, 2016	Qualitative study. Describes four different referral pathways in Scotland for transition to adult care.
Young, N. L., Gilbert, T. K., McCormick, A., Ayling-Campos, A., Boydell, K., Law, M., Fehlings, D. L., Mukherjee, S., Wedge, J. H., Williams, J. I., Youth and Young Adults With Cerebral Palsy: Their Use of Physician and Hospital Services, Archives of Physical Medicine and Rehabilitation, 88, 696-702, 2007	Reports mean rates of acute care admission and length of stay - but SDs are not reported so youth and adult groups cannot be compared.
Young, N. L., McCormick, A. M., Gilbert, T., Ayling-Campos, A., Burke, T., Fehlings, D., Wedge, J., Reasons for hospital admissions among youth and young adults with cerebral palsy, Archives of Physical Medicine and Rehabilitation, 92, 46-50, 2011	Describes reasons for (and length of) admission in adults with CP - but does not compare different service models.
Zwicker, J., Oskoui, M., Addressing heterogeneous needs using cerebral palsy registers, Developmental Medicine and Child Neurology, 59, 458-459, 2017	Commentary on another study (in children with CP)
Zwicker, J., Zaresani, A., Emery, J. C. H., Describing heterogeneity of unmet needs among adults with a developmental disability: An examination of the 2012 Canadian Survey on Disability, Research in Developmental Disabilities, 65, 1-11, 2017	Healthcare service models not compared

CP: cerebral palsy; HRQoL: health related quality of life; SD: standard deviation.

Economic studies

No economic evidence was identified for this review.

Appendix L – Research recommendations

Research recommendations for review question F1: What is the most clinical and cost effective configuration of services (setting and staffing) for adult with cerebral palsy?

No research recommendations were made for this review.