Report WP 2.3.2 Economic analysis

1. Introduction

This appendix focuses on the economic analysis conducted as part of the second work package. The main quantitative empirical part of the second work package is a four year longitudinal study of young people with diabetes, cerebral palsy or autistic spectrum disorder (ASD), who are aged 14-18 at baseline. These people are followed up three times, on a roughly annual basis, after being recruited at baseline. During each of these visits, the young people complete a battery of outcome measures. There are three sets of outcome measures that are most relevant to the economic component of the work:

1) The EQ-5D-Y. This is a generic health status measure developed for completion of people aged between 7+ and is an adaptation of the EQ-5D measure which is primarily aimed at adults. Since the programme has started the EQ-5D has been validated for people aged 12+. We have continued to you the EQ-5D-Y to remain consistent and the main difference between the two versions is how the EQ-5D-Y describes one aspect of daily living. Within the cohort study the EQ-5D-Y is completed at baseline and each of the 3 subsequent visits.

2) Health and social service contacts. At each visit a form was completed, describing the nature, type and number of contacts is collected. The young person also kept a diary of all the contacts they had had with the NHS since their previous visit. Further, data was obtained from the young person's medical notes and this was reviewed prior to the young person's visit. The resulting data (called the 'Final Account of Consultations) provides details of the use of services for each young person over the follow-up visit.

3) Stated preference exercise. The young people complete once during the third or fourth visit (depending upon whether they met the third scheduled visit) a discrete choice experiment (DCE) questionnaire.

These data were also collected to inform a proposed economic evaluation model. The design conduct and analysis of the discrete choice experiment are covered earlier in this appendix. We now cover: (1) the longitudinal data quality of life data; (2) the longitudinal cost data; and (3) the proposed economic model.

2. Methods

2.1. Longitudinal data analysis

The analysis of the 'economic' data as part of the longitudinal cohort followed similar principles to those used for the statistical analysis of outcome data reported for WP2.1. The dependent variables are health related quality of life as measured by the EQ-5D-Y and costs.

2.1.1 EQ-5D-Y

As noted already the EQ-5D-Y was collected 4 times within the longitudinal data set. The responses to the EQ-5D-Y were considered in two ways. First EQ-5D-Y data were converted into a utility score using the value set for adults.[103] Second they were summed with equal weighting to each of the five component questions. In this simple scoring system each of the five domains of the EQ-5D-Y was given equal weighting, and each level of each domain was scored 1, 2, or 3 depending on severity. This meant that each individual's response to the EQ-5D-Y could be scored between 5 and 15. This is defined hereafter as the sumscore. Lower scores by domain and sumscore indicate better health related quality of life.

These two approaches were adopted because there are known difficulties with applying the adult EQ-5D value set to the EQ-5D-Y, which does not have its own value set. However, it should also be noted that the study population was, on average, aged 16 or over at baseline and the adult EQ-5D questionnaire can be used with such a population. So, while the EuroQol Foundation note the EQ-5D-Y is a separate instrument, we expect that most of our study population would have been able to complete the adult version and, had they done so, it would be possible to apply utility values to such results without question.

Coupled with data on the young person's last date in paediatrics (considered the date of transfer), and also the visit (two, three or four) by which the young person had transferred, we calculated two definitions for their average pre- and post-transfer utility and their average pre- and post-transfer sumscore.

The plan was to assess the impact of the proposed beneficial features as well as demographic features of condition (diabetes, cerebral palsy or ASD), age at baseline, gender and transfer status (child services, adult services or general practice) using generalised linear modelling (GLM). This approach was adopted because of the highly skewed nature of sumscore, utility data. To conduct GLM fitting we utilised STATA and additional 'glmdiag' code [http://www.uphs.upenn.edu/dgimhsr/stat-cstanal.htm; accessed August 2017] and methods described by Glick and colleagues [http://www.uphs.upenn.edu/dgimhsr/documents/acadhlth.glick.061008.pdf accessed August 2017].

For utilities, there were no appropriate distributions (from either Gaussian, Inverse Gaussian, Gamma or Poisson) combined with tried link functions (cube root, square root, reciprocal, identity, log, squared, cubed) that could successfully model results. For sumscore the inverse Gaussian distribution with a log link and the gamma distribution with a log link were also considered plausible. Nevertheless, results from these analyses did not appear robust.

As a consequence an exploratory analysis estimating the median scores when a specific proposed beneficial feature was present were estimated.

2.1.2 *NHS, social service and costs to young people and their families*

The use of services all elicited as part of the longitudinal study were used in the estimation of costs. The costs data were then used as dependent variables in a set of regression analyses similar to those outlined above for the EQ-5D-Y data.

The perspective for the estimation of costs is the NHS and personal social services. Thus, it covers use of primary and secondary NHS care services, personal and social services and out-of-pocket expenses for the costs of accessingcare (time and travel).

NHS and personal social service costs

Use of NHS, personal and social services based upon each young person's Final Account of Consultations obtained at each visit. A unit cost was assigned to each item of resource use. These unit costs were sought from standard data sources such as the Unit Costs of Care for, for example, general practitioner visits.

The unit cost of medications was taken from the British National Formulary (BNF) and unit cost of inpatient and/or day case stays was derived from NHS reference costs. As insulin prescriptions were not consistently mentioned in the data, these were costed separately according to existing literature sources[209, 210] inflated to the current price year (2016) and whether or not the young person was documented as receiving an insulin pump in the Final Account notes. Visits that were cancelled in advance did not incur costs, but those where either the respondent or the staff member did not attend, incurred 50% of the cost of a typical visit of that type.

Once all documented resource use items received a unit cost, data were combined with information on each participant's transfer status to calculate average pre- and post-transfer costs across all participants.

The plan was to assess the impact on cost of the proposed beneficial features as well as demographic features of condition (diabetes, cerebral palsy or ASD), age at baseline, gender and transfer status (child services, adult services or general practice) using generalised linear modelling (GLM). A similar problem to that described above was encountered and hence an exploratory analysis estimating the median scores when a specific proposed beneficial feature was present were estimated.

2.2. Economic evaluation modelling

The purpose of the economic modelling was to compare the costs and effects of alternative hypothetical ways (termed interventions in the economic model) of delivering care during transition. The different transitions interventions were to be defined in terms of the presence or absence of the potential beneficial features. Data on the impact of the potential beneficial features was to come from the longitudinal analysis described in WPs 2.1 and 2.2. A model such as this however has limited value unless it is plausible that the potentially beneficial features could influence costs and effects. Therefore, its conduct was conditional on the findings of WPs 2.1 and 2.2.

Figure 17 gives the structure of the model. In the model all young people start in child services and are aged 14 years old and are followed for 10 years; the time over which a young person would be expected to be undergoing transition. The model considers a hypothetical cohort of 1,000 young people. Each year they have a probability of transferring out of child services and being discharged into primary care or moving into adults services. If in any given year they do not make the transfer then they remain in child services indicated by the ellipse connected to the Child Services state. In each state an annual cost, utility or sumscore value was assigned. The values used for this were derived from the work conducted under WPs 2.1 and 2.2. By combining these values with the chance of being in a state in give year it was possible to estimate cumulative costs, cumulative utilities (which because they are estimated over time provide an estimate of quality adjusted life years – QALYs) and sumscore.

Figure 17: Structure of the economics model
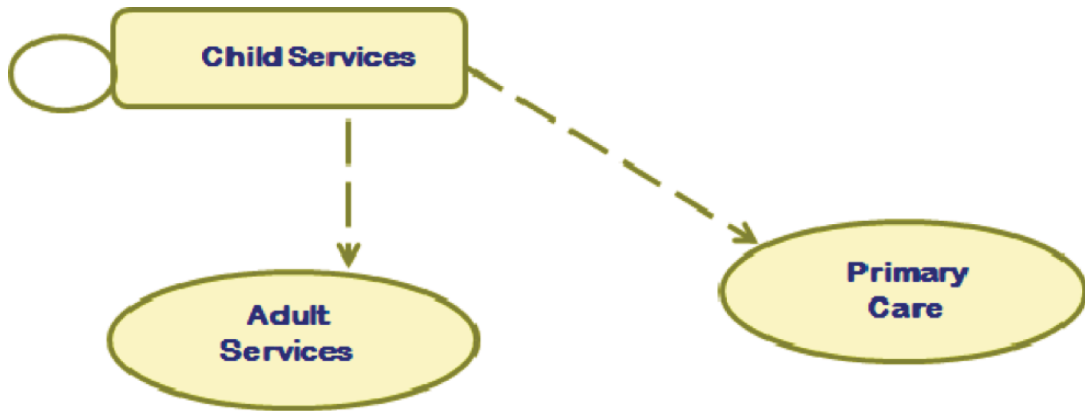
The chance of being in a given state in a given year was given by the rate of transfer. The rate of transfer was calculated by considering the number of people who transferred from child services each year, and the ratio of transferred young people who were transferred to adult services compared with PRIMARY CARE services. This decline in the proportion of young people in child services is modelled by the exponential distribution whereby $y = n(e^{-0.498t})$, where n is the baseline sample size and t is the number of years since baseline. Of those who transfer, approximately 55% go to adult services, with the remainder of those who transferred going to primary care.

The cumulative effects were estimated for hypothetical transition services. The first of these was a service that did not contain any of the proposed beneficial features. Alternative services were defined in terms of the presence of a single proposed beneficial features, thus there was an alternative service was defined for each of the nine proposed beneficial features.

The difference in cost and QALYs and sumscore between each alternative service and a service containing no proposed beneficial features. These data were then used to estimate the extra cost required to produce one more unit of effect – the incremental cost-effectiveness ratio (ICER). All costs are presented in terms of a common price year (2016) and all costs and consequences are discounted at 3.5%.

(Discounting is used because the when a cost and benefit occurs is believed to be important. It reflects the fact that costs and benefits that occur in the future are given less weight than costs and benefits that occur now. 'This time preference' for when costs and benefits might occur is captured by the use of a discount rate, which reduces the importance of future costs and benefit in an analysis.)

The presence or absence of the proposed beneficial features was elicited in two ways. A form was completed each year by the research associate from data in the medical notes. It consisted of details of clinic visits, professionals in attendance, medical tests undertaken and whether PBFs had been experienced. This was called the 'Final Account of Consultations'. Following the home visits with further discussion of appointments and PBFs and completion with the young person of a questionnaire, it became clear that the Final Account was incomplete for some young people. Therefore analysis was also undertaken with data combined from both sources.

2.3. Balance sheet analysis

The results of the model were combined with finding from other areas of the report in a balance sheet analysis. In this analysis we have presented those outcomes that could favour a hypothetical transition intervention compared to a situation where there is no support during the transition from child to adult services. The approach can incorporate quantitative data such as that produced from the statistical analysis of the longitudinal study, evidence from the broader literature and evidence from the other work conducted within the programme.

3. Results

3.1. Longitudinal data analysis

3.1.1. EQ-5D results

Of the 375 participants in the Transition Research Programme, 66 left the study. EQ-5D-Y data were available for 373 respondents at baseline, and 244 respondents by visit 4.

Summary response data for the EQ-5D-Y

For each of the five domains, the minimum and maximum scores were the extremes of the 3-levels (1 and 3 respectively, and the median score was consistently 1, meaning that for every domain at every time point, at least half the respondents reported 'no problems'. The number and proportion of respondents responding at each of the three levels for each of the five domains is provided in Table 28. For most domains the proportion of respondents report some or lots of problems (scores 2 and 3) increase over time. The mean score also suggest that health may have declined over time (score look to have increased on average for all domains except self-care).

Table 28: Responses by each of the domains of the EQ-5D-Y over the follow-up

		Scoring 1		Scoring 2		Scoring 3		Missing		Mean Score	SD
		n	%	n	%	n	%	n	%		
Mobility	Baseline	298	79.5%	63	16.8%	12	3.2%	2	0.5%	1.23	0.49
	Visit 2	239	63.7%	59	15.7%	6	1.6%	71	18.9%	1.23	0.47
	Visit 3	190	50.7%	52	13.9%	14	3.7%	119	31.7%	1.31	0.57
	Visit 4	189	50.4%	44	11.7%	11	2.9%	131	34.9%	1.27	0.54
Self-care	Baseline	311	82.9%	50	13.3%	12	3.2%	2	0.5%	1.20	0.47
	Visit 2	258	68.8%	35	9.3%	11	2.9%	71	18.9%	1.19	0.47
	Visit 3	214	57.1%	30	8.0%	11	2.9%	120	32.0%	1.20	0.50
	Visit 4	204	54.4%	30	8.0%	10	2.7%	131	34.9%	1.20	0.50
Usual Activities	Baseline	273	72.8%	96	25.6%	4	1.1%	2	0.5%	1.28	0.47
	Visit 2	226	60.3%	73	19.5%	5	1.3%	71	18.9%	1.27	0.48
	Visit 3	163	43.5%	90	24.0%	3	0.8%	119	31.7%	1.38	0.51
	Visit 4	170	45.3%	70	18.7%	4	1.1%	131	34.9%	1.32	0.50
Pain	Baseline	245	65.3%	122	32.5%	6	1.6%	2	0.5%	1.36	0.51
	Visit 2	200	53.3%	95	25.3%	9	2.4%	71	18.9%	1.37	0.54
	Visit 3	163	43.5%	88	23.5%	5	1.3%	119	31.7%	1.38	0.53
	Visit 4	154	41.1%	78	20.8%	12	3.2%	131	34.9%	1.42	0.59
Anxiety	Baseline	236	62.9%	117	31.2%	20	5.3%	2	0.5%	1.42	0.59
	Visit 2	177	47.2%	106	28.3%	21	5.6%	71	18.9%	1.49	0.62
	Visit 3	142	37.9%	95	25.3%	19	5.1%	119	31.7%	1.52	0.63
	Visit 4	125	33.3%	102	27.2%	17	4.5%	131	34.9%	1.56	0.62

The variations in EQ-5D-Y responses by the three conditions is reported in Table 29. As this table show that young people with diabetes consistently experienced fewest problems in any domain. Those with cerebral palsy were more likely to experience problems in the mobility and self-care domains than young people with diabetes or ASD. The proportion of respondents with ASD reporting problems in the anxiety domain was consistently higher than for the other two conditions. As these differences were present at baseline, they may simply reflect the nature of the conditions under consideration.

Table 29: Summary of EQ-5D-Y responses by condition

Dimension	Time point	% experiencing problems (score 2 or 3) including missing			% experiencing problems (score 2 or 3) excluding missing		
		ASD	CP	Diabetes	ASD	CP	Diabetes
Mobility	Baseline	16.1%	47.7%	3.3%	16.2%	48.1%	3.3%
	Visit 2	11.9%	40.2%	5.3%	15.4%	50.6%	6.3%
	Visit 3	14.4%	43.0%	2.0%	20.5%	63.0%	3.0%
	Visit 4	10.2%	36.4%	2.7%	15.8%	56.5%	4.0%
Self-care	Baseline	15.3%	40.2%	0.7%	15.4%	40.6%	0.7%
	Visit 2	8.5%	31.8%	1.3%	11.0%	40.0%	1.6%
	Visit 3	11.9%	24.3%	0.7%	16.9%	35.6%	1.0%
	Visit 4	10.2%	26.2%	0.0%	15.8%	40.6%	0.0%
Usual activities	Baseline	36.4%	37.4%	11.3%	36.8%	37.7%	11.3%
	Visit 2	24.6%	29.0%	12.0%	31.9%	36.5%	14.1%
	Visit 3	35.6%	32.7%	10.7%	50.6%	47.9%	16.0%
	Visit 4	26.3%	25.2%	10.7%	40.8%	39.1%	16.2%
Pain	Baseline	38.1%	51.4%	18.7%	38.5%	51.9%	18.7%
	Visit 2	27.1%	42.1%	18.0%	35.2%	52.9%	21.1%
	Visit 3	29.7%	37.4%	12.0%	42.2%	54.8%	18.0%
	Visit 4	25.4%	34.6%	15.3%	39.5%	53.6%	23.2%
Anxiety	Baseline	65.3%	28.0%	20.0%	65.8%	28.3%	20.0%
	Visit 2	50.8%	29.9%	23.3%	65.9%	37.6%	27.3%
	Visit 3	46.6%	27.1%	20.0%	66.3%	39.7%	30.0%
	Visit 4	46.6%	27.1%	23.3%	72.4%	42.0%	35.4%

ASD = Autism spectrum disorder; CP = cerebral palsy

EQ-5D Sumscore
The higher the sumscore the worse health is reported to be. The median sum score for all participants across all time points was 6 (IQR: 2.8), which indicates at least half of participants did not experience 'no problems' across all domains (a score of 5 is required). Average sumscores were not associated with gender, nor were pre or post transfer scores. In addition, there were significant differences in both sumscores by condition, as diabetes participants had significantly lower scores (i.e. better health) ($p<0.001$) at all time points (Table 30).

EQ-5D-Y Utilities
The median utility score across all participants was 0.83 (IQR: 0.304). Average utility scores were not associated with gender nor were pre or post transfer scores. However, among those who transferred, scores were significantly higher (Wilcoxon Signed Rank test 7465.5, $p<0.001$) for those doing so to adult services (median 0.86, IQR: 0.37) compared to Primary Care services (median 0.78, IQR: 0.38). As was found for the sumscore participants with diabetes had a higher significantly higher scores ($p<0.001$) at all time points (Table 30).

Table 30: Summary Sumscore and Utility score by condition pre- and post-transfer

	All (IQR)	ASD (IQR)	Cerebral palsy (IQR)	Diabetes (IQR)
n	301	96	83	122
Median sum score	6 (2.79)	7 (2)	7 (2.98)	5.2 (1)
Median utility score	0.83 (0.30)	0.75 (0.27)	0.70 (0.43)	0.94 (0.14)
n	269	86	70	113
Median pre-transfer sumscore	6 (2)	7 (2)	7 (3)	5 (1)
Median pre-transfer utility score	0.85 (0.27)	0.75 (0.26)	0.72 (0.43)	0.95 (0.13)
n	230	68	66	96
Median post-transfer sumscore	6 (3)	7 (2)	7 (2.75)	5 (1)
Median post transfer utility score	0.82 (0.29)	0.76 (0.24)	0.70 (0.33)	0.95 (0.16)

IQR = Interquartile range; ASD = Autism spectrum disorder

3.1.2   Use of services and costs
Use of services overall and by condition

The number of participants providing data was 286 at visit 2, 249 at visit 3 and 216 at visit 4, representing 76.2%, 66.4% and 57.6% of the total sample respectively. The young people in the sample recorded a total of 8383 visits over the course of the follow-up. Of these 7755 (92.5%) occurred, with others either being cancelled in advance or without sufficient prior notification (i.e. 'did-not-attend' or 'DNA'd'). Corresponding to the fall in the sample contributing data the number of visits occurring at each time point declined from 3293 (42.5% of all visits) at visit 2, to 2476 (31.9% of all visits) at visit 3 and 1986 visits (25.6% of all visits) at visit 4.

The average number of visits experienced by the participants declined from a median of 7 (IQR 8) at visit 2, to a median of 6 (IQR 9) at visit 3 and 4 (IQR 9) at visit 4. The median number of visits across the whole duration of follow-up was 19 (IQR 20), but this varied depending on condition, ranging from 14 (IQR: 27.8) for those with cerebral palsy to 23 visits for those with diabetes (IQR: 13.8). Those participants with ASD had a median of 16 visits (IQR: 23).

Over time, the median number of visits, among participants who experienced visits over the time period decreased for those with cerebral palsy and diabetes (from 7 and 8 at visit 2 respectively to 5 and 7 at visit 4), whereas for those with ASD the median number of visits increased from 5 to 6 over the same time period. If we include those who had no visits here, this trend is lost (cerebral palsy goes from 6.5 to 1 and Diabetes goes from 8 to 6, ASD goes from 4 to 3). The composition of visits also varied in terms of venue and staffing, both across time and for each of the three conditions (Table 31).

As Table 31 illustrates for those with ASD, visits were typically held in community/high street venues (this includes CAMHS) or home/school visits. Whereas for those with cerebral palsy visits typically took place at either home/school or outpatient clinics. Over half of contacts for those with diabetes took place in outpatient settings at each visit, and the majority of 'other' types of contact were comprised of telephone conversations with clinicians.

Table 32 illustrates the type of staff seen and as this table shows there was a very small number of multidisciplinary contacts. Compared to participants with diabetes, participants with ASD and cerebral palsy were less likely to have multidisciplinary visits, although the proportion of visits that were multi-disciplinary for those with ASD increased over time, whereas for those with diabetes they decreased. The proportion of visits involving doctors increased over time for those with diabetes, whereas it declined for those with ASD. Visits involving other care providers (typically social care staff or volunteers working with the young person) increased in those with ASD as did nursing visits, whereas the latter decreased for those with diabetes.

Table 31: Percentage of all health care contacts by provider and by condition

	% of all visits of this type at visit 2	% of all visits of this type at visit 3	% of all visits of this type at visit 4
All			
Community	17.5%	16.8%	12.6%
Primary care	7.0%	8.9%	12.5%
Home visit	34.5%	29.7%	26.0%
Inpatients (elective)	0.6%	0.6%	0.6%
Inpatients (emergency)	0.9%	1.2%	1.3%
Outpatients	30.2%	33.9%	39.1%
Other	9.5%	8.8%	7.9%
ASD			
Community	53.1%	29.2%	22.3%
Primary care	5.6%	11.2%	17.6%
Home visit	25.7%	43.9%	41.7%
Inpatients (elective)	0.0%	0.1%	0.3%
Inpatients (emergency)	0.3%	1.5%	1.4%
Outpatients	3.4%	7.7%	10.4%
Other	11.8%	6.4%	6.3%
Cerebral palsy			
Community	6.5%	17.4%	15.5%
Primary care	4.5%	4.0%	4.7%
Home visit	63.1%	46.8%	39.9%
Inpatients (elective)	0.5%	0.8%	1.1%
Inpatients (emergency)	1.0%	0.5%	0.9%
Outpatients	23.6%	28.9%	34.8%
Other	0.8%	1.5%	3.0%
Diabetes			
Community	2.0%	4.5%	1.8%
Primary care	10.6%	10.4%	14.3%
Home visit	10.5%	3.8%	1.3%
Inpatients (elective)	1.1%	0.9%	0.6%
Inpatients (emergency)	1.4%	1.5%	1.6%
Outpatients	57.8%	62.4%	67.2%
Other	16.5%	16.6%	13.1%

ASD = autism spectrum disorder

Table 32: Descriptive statistics on use of health services

All	From baseline to visit 2	From visit 2 to visit 3	From visit 3 to visit 4
% healthcare contacts with doctor(s) present	31.9%	33.4%	32.6%
% healthcare contacts with nurse(s) present	28.3%	26.4%	26.1%
% healthcare contacts with AHP*(s) present	38.2%	34.2%	28.6%
% healthcare contacts with others present	16.4%	18.8%	24.7%
% of healthcare contacts: multidisciplinary	13.1%	13.0%	11.5%
Diabetes	Visit 2	Visit 3	Visit 4
% healthcare contacts with doctor(s) present	39.4%	41.6%	43.6%
% healthcare contacts with nurse(s) present	66.3%	56.6%	51.7%
% healthcare contacts with AHP(s) present	29.2%	29.7%	29.2%
% healthcare contacts with others present	2.1%	1.9%	1.7%
% of healthcare contacts : multidisciplinary	32.0%	29.2%	24.4%
Cerebral palsy	Visit 2	Visit 3	Visit 4
% healthcare contacts with doctor(s) present	18.6%	23.1%	20.1%
% healthcare contacts with nurse(s) present	3.8%	2.6%	2.9%
% healthcare contacts with AHP(s) present	56.6%	36.5%	41.2%
% healthcare contacts with others present	23.4%	39.5%	38.0%
% of healthcare contacts: multidisciplinary	2.6%	2.0%	2.7%
Autism spectrum disorder	Visit 2	Visit 3	Visit 4
% healthcare contacts with doctor(s) present	40.6%	32.9%	31.3%
% healthcare contacts with nurse(s) present	13.0%	12.9%	18.0%
% healthcare contacts with AHP(s) present	23.9%	37.1%	16.1%
% healthcare contacts with others present	25.4%	20.6%	39.0%
% of healthcare contacts: multidisciplinary	3.0%	4.4%	4.6%
*AHP Allied Health Professional			

Combining data from Table 31 with unit cost data allowed a cost of care for each young person to be estimated. Data were available on 301 young people (78.6%). The overall median cost per participant was £4954 (IQR: £6103). Diabetes was the most costly condition, and cerebral palsy was the least costly although there was considerable variation between individuals. The difference between the cost of those with diabetes and those with ASD was statistically significant (p<0.001). The difference in pre and post transfer costs was not statistically significant at the 0.05 level although there was a trend both overall and by each condition (Table 33).

Table 33: Estimated median costs, by condition both overall and pre and post transfer

	All (IQR)	ASD (IQR)	Cerebral palsy (IQR)	Diabetes (IQR)
N	301	96	77	125
Median total costs	£4954 (£6103)	£2351 (£4211)	£2394 (£5606)	£7003 (£3667)
Median total costs per annum	£2307 (£2705)	£1659. (£2573)	£1447 (£2725)	£2915 (£2422)
N	279	88	67	121
Median **pre-transfer** costs	£2420 (£4506)	£1694 (£2454)	£1370 (£4215)	£4434 (£5136)
Median **pre-transfer** costs per annum	£1673 (£2242)	£915 (£1277)	£949 (£1974)	£2410 (£1587)
N	194	56	48	87
Median **post-transfer** costs	£2080 (£3470)	£1262 (£2540)	£1395 (£3015)	£3114 (£3440)
Median **post-transfer** costs per annum	£1371 (£1862)	£645 (£1883)	£707. (£1673)	£1849 (£1193)

Presence of proposed beneficial features

The presence or absence of the proposed beneficial features was elicited in two ways.

A form was completed each year by the research associate from data in the medical notes. It consisted of details of clinic visits, professionals in attendance, medical tests undertaken and whether PBFs had been experienced. This was called the 'Final Account of Consultations'. Following the home visits with further discussion of appointments and PBFs and completion with the young person of a questionnaire, it became clear that the Final Account was incomplete for some young people. Therefore analysis is also undertaken with data combined from both sources.

Sensitivity analyses were carried using these alternative definitions.

Details regarding the nine criteria used to define the amount of PBF required to be 'satisfactory' is defined elsewhere. When this was applied to the data, the following numbers of young people met the criteria for receiving each PBF – Table 34. This has been shown by the two data sources. These young people did not always also have available cost data, but where they did have cost data this is reported in Table 34 in parenthesis.

3.2 Economic evaluation model

Using the data reported above in the proposed economic model an attempt was made to estimate the impact of the proposed beneficial features on sumscores, utility score, QALYs and costs. The model sought to reflect the fact that the young person's place of care would change over time as they leave child services. On leaving child services the young person would be either receive care in primary care or secondary care.

Table 35 shows the input parameters for costs, utilities and sumscore. The data reported in Table 35 are point estimates and whilst there may be apparent trend in the data it is important to remember that confidence intervals around these values would be very wide. Therefore, for the purposes of making decisions about care their quality would be judged as low. However, when interpreted with other findings they may help highlight areas for further research.

Table 34: Number of young people experiencing the proposed beneficial features when experience is captured in different ways

	All (for whom cost data are available)	Child services (for whom cost data are available)	Transferred to adults services (for whom cost data are available)	Transferred to primary care (for whom cost data are available)	Left study
As captured from Final Account					
No PBFs	35 (33)	9 (9)	11 (11)	15 (13)	0
Meet adult team before transfer	66 (60)	3 (3)	57 (52)	5 (5)	1
Age-banded clinic	141 (129)	25 (25)	108 (97)	8 (7)	0
Holistic life-skills training	128 (114)	29 (29)	80 (73)	18 (12)	1
Key worker	58 (50)	12 (12)	37 (32)	9 (6)	0
Transition manager for clinical team	23 (21)	3 (3)	17 (16)	2 (2)	1
Appropriate parent involvement	11 (11)	3 (3)	8 (8)	0 (0)	0
Promotion of health self-efficacy	30 (27)	4 (4)	26 (23)	0 (0)	0
Coordinated team	95 (84)	27 (27)	53 (48)	14 (9)	1
Transition plan	26 (24)	5 (5)	20 (19)	0 (0)	1
All PBFs	0 (0)	0 (0)	0 (0)	0 (0)	0
As captured from both sources (face-to-face contacts and Final Account)					
No PBFs	7 (7)	1 (1)	2 (2)	4 (4)	0
Meet adult team before transfer	121 (108)	12 (12)	91 (84)	15 (12)	3
Age-banded clinic	163 (142)	26 (26)	119 (107)	13 (9)	5
Holistic life-skills training	149 (129)	31 (31)	91 (83)	23 (15)	4
Key worker	78 (66)	17 (17)	54 (45)	7 (4)	0
Transition manager for clinical team	70 (61)	13 (13)	45 (44)	7 (4)	5
Appropriate parent involvement	121 (108)	18 (18)	73 (69)	30 (21)	0
Promotion of health self-efficacy	100 (90)	19 (19)	70 (63)	11 (8)	0
Coordinated team	195 (170)	45 (45)	122 (111)	21 (14)	7
Transition plan	56 (50)	9 (9)	41 (39)	2 (2)	4
All PBFs	1 (1)	1 (1)	0 (0)	0 (0)	0

Table 35: Costs, utilities and sumscore data   Cost

	Costs											
	PBFs captured from final account						PBFs captured from final account and face-to-face					
State	Child services		Transferred adult services		Transferred primary care		Child services		Transferred adult services		Transferred primary care	
	Median	IQR	Median	IQR	Median	IQR	Median	IQR	Median	IQR	Median	IQR
No PBFs	816	1,162	800	1,247	662	831	658	-	1,245	1,027	199	336
Meet adult team before transfer	2,915	1,554	1,823	1,222	845	699	3,188	1,793	1,761	1,213	754	1,043
Age-banded clinic	2,915	1,837	1,894	1,263	2,325	764	2,477	1,576	1,735	1,347	2,325	976
Holistic life-skills training	2,456	2,328	1,708	1,609	569	2,114	2,616	2,254	1,677	1,666	504	2,206
Key worker	3,233	2,727	1,546	1,356	2,431	1,519	2,497	2,179	1,600	1,376	731	1,087
Transition manager	2,616	4,777	1,644	2,963	5,114	2,576	2,616	1,527	1,510	1,738	1,623	3,292
Appropriate parent involvement	NA	1,373	NA	2,447	NA	NA	2,400	2,760	1,487	1,929	566	669
Promotion of health self-efficacy	NA	1,616	NA	1,232	NA	NA	2,794	1,892	1,600	1,362	401	1,267
Coordinated team	2,915	2,712	1,350	1,626	853	2,002	2,725	1,633	1,677	1,498	1,040	1,933
Transition plan	NA	1,258	NA	2,026	NA	NA	3,391	1,258	1,894	1,817	1,136	582
All PBFs	NA	NA	NA	NA	NA	NA	NA	-	NA	NA	NA	NA

Table 35: Costs, utilities and sumscore data   Utilities

	Utility scores											
	PBFs captured from final account						PBFs captured from final account and face-to-face					
State	Child services		Transferred adult services		Transferred primary care		Child services		Transferred adult services		Transferred primary care	
	Median	IQR	Median	IQR	Median	IQR	Median	IQR	Median	IQR	Median	IQR
No PBFs	0.90	0.19	0.66	0.47	0.80	0.26	0.90	0.00	0.41	0.34	0.87	0.18
Meet adult team before transfer	0.36	0.44	0.98	0.17	0.83	0.09	0.82	0.45	0.92	0.22	0.80	0.23
Age-banded clinic	0.88	0.16	0.91	0.22	0.64	0.32	0.87	0.15	0.91	0.24	0.61	0.19
Holistic life-skills training	0.83	0.40	0.89	0.26	0.81	0.12	0.83	0.35	0.88	0.30	0.81	0.12
Key worker	0.87	0.19	0.92	0.21	0.82	0.12	0.86	0.22	0.92	0.24	0.82	0.17
Transition manager	0.73	0.28	0.77	0.42	0.92	0.06	0.83	0.44	0.81	0.26	0.73	0.53
Appropriate parent involvement	NA	0.30	NA	0.28	NA	NA	0.79	0.45	0.85	0.33	0.78	0.20
Promotion of health self-efficacy	NA	0.06	NA	0.18	NA	NA	0.87	0.18	0.91	0.19	0.80	0.22
Coordinated team	0.87	0.52	0.82	0.28	0.68	0.30	0.86	0.22	0.90	0.25	0.68	0.29
Transition plan	NA	0.12	NA	0.15	NA	NA	0.88	0.51	0.85	0.22	0.75	0.07
All PBFs	NA	NA	NA	NA	NA	NA	NA	0.00	NA	NA	NA	NA

Table 35: Costs, utilities and sumscore data   Sumscores

Sumscores												
	PBFs captured from final account						PBFs captured from final account and face-to-face					
State	Child services		Transferred adult services		Transferred primary care		Child services		Transferred adult services		Transferred primary care	
	Median	IQR	Median	IQR	Median	IQR	Median	IQR	Median	IQR	Median	IQR
No PBFs	6.00	2	8.00	3	7.00	2	6.00	0	9.50	2.5	6.00	0.5
Meet adult team before transfer	10.00	2.5	5.00	1	6.50	1.25	6.00	4	6.00	2	7.00	2
Age-banded clinic	6.00	1.5	6.00	2	8.00	1.75	6.00	1	6.00	2	8.00	1
Holistic life-skills training	6.00	2	6.00	2	6.50	1	6.00	2	6.00	2	6.50	1
Key worker	6.00	1	6.00	1.5	7.00	1	6.00	2	5.00	2	7.00	1
Transition manager	7.00	2	7.00	3	5.50	0.5	6.00	3	6.00	2	7.00	2.75
Appropriate parent involvement	NA	1.5	NA	2	NA	NA	6.50	3	6.00	3	7.00	2
Promotion of health self-efficacy	NA	0.25	NA	1	NA	NA	6.00	1.5	6.00	2	7.00	2
Coordinated team	6.00	4	6.00	3	8.00	2	6.00	2	6.00	2	7.50	2
Transition plan	NA	1	NA	2	NA	NA	6.00	4	6.00	2	7.00	1
All PBFs	NA	NA	NA	NA	NA	NA	NA	0	NA	NA	NA	NA

Using these data within the economic model described in Section 2.2 and modelling over a 10 year time horizon it is possible to estimate how costs, sumscore and QALYs vary by presence or absence of proposed beneficial feature. This analysis considers each proposed beneficial feature separately and given the limited data available as reported in Table 34 should be considered as exploratory only. Given the exploratory nature probabilistic sensitivity analysis has not been conducted as there is considerable imprecision in result. The results are best considered as either illustrative of the approach take or hypothesis generating about possible proposed beneficial features to consider when developing an intervention to improve transition.

Table 36 shows estimated costs, effects (sumscore and QALY) and Table 37 the incremental cost-effectiveness for the comparison of a service incorporating each proposed beneficial feature alone a service where no features were present. All outcomes are discounted (each at 3.5%) It was not possible to compare individual PBFs against a situation where all PBFs were provided, as only one young person experienced all PBFs in the study. As the presence of the proposed beneficial features is based on reports during face to face contacts a sensitivity analysis has been performed using these data.

The data reported in Tables 36 and 37 illustrate that the results are sensitive to the methods used to define whether a proposed beneficial feature was present. In general there were more reports of the PBFs being present. The higher the incremental cost-effectiveness ratio the less likely an intervention would be considered value for money.

In relative terms there were several PBFs that seemed to perform better regardless of whether the definition of 'satisfactory' PBF exposure is based on final account or face-to-face data. These were the provision of holistic care; having a key worker, and, (where the sample was sufficient to quantify it) promotion of health self-efficacy.

Meeting the adult team was sensitive to the source of data and was considerably better when estimates were based on face to face data. Having a written transition plan was one of the poorer performing proposed beneficial features as was having a Transition manager for clinical team.

Table 36: Estimated costs and effects for each intervention

PBFs captured from Final Account of Consultations			
	Mean costs of strategy over model time horizon	Mean QALYs for strategy over time horizon	Mean sumscore for strategy over model time horizon
No PBFs	£6552	6.65	8
Meet adult team before transfer	£15,726	6.48	8
Age-banded clinic	£20,018	7.02	8
Holistic life-skills training	£13,406	7.28	7
Key worker	£19,922	7.53	7
Transition manager	£26,134	6.96	8
Appropriate parent involvement	NA	NA	NA
Promotion of health self-efficacy	NA	NA	NA
Coordinated team	£14,116	6.80	8
Transition plan	NA	NA	NA
All PBFs	NA	NA	NA
PBFs captured using responses to Final Account and face to face questionnaire			
No PBFs	6415	6.02	9
Meet adult team before transfer	15,941	7.36	7
Age-banded clinic	18,396	6.93	8
Holistic life-skills training	13,519	7.28	7
Key worker	13,592	7.51	7
Transition manager	16,042	6.78	7
Appropriate parent involvement	12,912	6.94	8
Promotion of health self-efficacy	13,644	7.43	7
Coordinated team	15,268	7.05	8
Transition plan	18,523	7.09	7
All PBFs	NA	NA	NA

Table 37: Incremental cost effectiveness

Cost per QALY gained	ICER using PBFs captured from final account	ICER using PBFs captured from final account and face to face questionnaire
Meet adult team before transfer	Dominated*	7121
Age-banded clinic	36,487	13,176
Holistic life-skills training	10,991	5618
Key worker	15,259	4811
Transition manager	64,359	12,592
Appropriate parent involvement	NA	7070
Promotion of health self-efficacy	NA	5127
Coordinated team	52,100	8596
Transition plan	NA	11,259
Cost per 1 unit change in sumscore		
Meet adult team before transfer	15,092	7011
Age-banded clinic	22,693	12,662
Holistic life-skills training	5692	4563
Key worker	13,363	3878
Transition manager	23,364	7114
Appropriate parent involvement	NA	5345
Promotion of health self-efficacy	NA	5341
Coordinated team	12,746	7700
Transition plan	NA	8947

* PBF on average more costly and less effective. Therefore it is dominated by a service without that PBF
ICER Incremental cost effectiveness ratio
QALY Quality adjusted life year
PBF Proposed beneficial feature

3.3   Balance sheet

Taking the data from the all the economic analyses and elsewhere within the programme a balance sheet summarising the pros and cons of developing a service which has some of the proposed beneficial features compared to a service without these features can be developed (Table 38). The purpose of Table 38 is to inform a decision about whether the cost of implementing a new service would be offset by any reductions in cost in the future and improvements in health.

The development and implementation of a service containing one or more proposed beneficial features would incur a cost, the magnitude of that cost will be determined by how that proposed beneficial feature is delivered. The limited data available from the economic model suggests that the implementation of a new service will increase the use of NHS services at least over a 10 year time horizon for a young person aged 14 entering a 'transition' service but there are no obvious harms to the young person of any of the proposed beneficial features.

The question for the decision-maker is whether the development of a flexible service capable of meeting the changing needs of young people, allowing appropriate parental involvement, promoting self-efficacy and more general independence of the young person would be worth these extra costs given that they may increase engagement with health services and prevent deterioration/exacerbation in health in the long-term.

Table 38: Pros and cons of a transition service containing some of the proposed beneficial features

Favours service containing the adaptability needed from the findings of the Q-sort, the preferences in the DCE and the proposed beneficial features in the prospective study.	Favours service not containing the adaptability needed from the findings of the Q-sort, the preferences in the DCE and the proposed beneficial features in the prospective study.	Source
	Avoided the currently unknown costs of providing beneficial features. Costs of some may be modest e.g. a 'written transition plan' whilst others are substantial e.g. adoption of a multi-disciplinary team.	Costs not estimated
A service should be flexible enough to meet the need of individuals – one size does not fit all and needs may change over time. Service needs to consider the condition, the stage of development, and the young person's personality.		Q-sort (WP 1.3) and DCE (WP 2.3.1)
	High predicted uptake of a service with no proposed beneficial features (78%; 95% CI 75-81)	DCE (WP 2.3.1)
Predicted uptake of a service higher when the service included parent involvement; a service where the same staff are seen at each clinic appointment; where the young receive good communication and are offered the opportunity to make decisions. - Increasing engagement may help prevent deterioration in the young person's condition over time	From the DCE a strong preference for current care; no strong preference for a 'key worker' or flexibility of appointments	DCE (WP 2.3.1) Anticipated impact not measured in study
'Appropriate parent involvement', 'Promotion of health self-efficacy', 'Meeting the adult team' improved outcomes.		Analysis of longitudinal data (WP 2.1)
Estimated additional benefit to health service of adopting any of the above features, except care by a co-ordinated team	Estimated additional cost to health services of adopting any of the proposed beneficial features	Economic model analysis (WP 2.3.2)
Longer term model cautiously suggests 'holistic life-skills training'; 'having a key worker', and 'promotion of health self-efficacy' might be value for money.	Longer term model cautiously suggests that a 'transition manager for clinical team', 'age-banded clinic' or 'meeting the adult team before transfer' might not be value for money'.	Economic model analysis (WP 2.3.2)
No evidence of a difference		
Lack of clarity over what was meant by a health passport, which might be valued but rarely used		Works conducted by UP (WP 1.2)
During transition, no evidence of any effect on outcomes of 'having a transition plan', 'attending an age-banded clinic', 'being cared for by a coordinated team'; 'being part of a service with a transition manager for clinical team'; 'having access to holistic life-skills training'		Analysis of longitudinal data (WP 2.1)
Over the duration of the longitudinal study, no evidence of any effect on costs or health related quality of life of 'having a transition plan', 'attending an age-banded clinic', 'being cared for by a co-ordinated team'; 'being part of a service with a Transition manager for clinical team'; 'having access to holistic life-skills training'.		Analysis of costs and EQ-5D data in the economic analysis (WP 2.3.2)

4 Limitations and strengths

Loss to follow-up may have led to biased estimates if those lost to follow-up differed systematically from those who were not. For this reason, we did not conduct an analysis of completers only (i.e. those for whom data at all four time points was available). On the factors for which we had data, there was no selective attrition except for a suggestion in those with cerebral palsy that more socio-economically deprived young people were likely to drop out.

A second concern was whether the HRQoL instrument was sufficiently sensitive to capture an influence of PBFs. The study was not powered on the EQ-5D-Y but rather on the outcomes used in *WP 2.1*. We did have sufficient power to identify clinically significant changes in the EQ-5D-Y but, for analysis by condition, sample size meant that some real effects of difference may not have been detectable. In relation to the PBFs, power was limited if a feature was often absent.

Alternatively, the HRQoL instrument might have been sufficiently sensitive but follow-up time was too short to capture any influence of PBFs.

The DCE suggested that experiencing some of the PBFs might increase young people's engagement with health services, which in turn might alleviate longer term harms (and costs) due to inadequate healthcare. The economic evaluation was exploratory and only compared services with individual PBFs to a service with no PBFs. Therefore, no probabilistic sensitivity analysis was conducted as is normally recommended for health technology assessment type economic evaluations and a quantified estimate of the effect associated with various combinations of PBFs was not possible. Therefore the economic conclusions should be interpreted with care.

It was necessary to annuitise the cost data for participants to compare with the EQ-5D-Y data. Thus, certain costs for participants for items mentioned occasionally but likely to apply over the course of the study were calculated as a daily rate following aggregation of the visit data. For example, as insulin prescriptions were not consistently reported in the data, these were costed separately according to existing literature and then inflated to the current price year. We think this made best use of the available data and is unlikely to have distorted the results.

The data collected on outcomes, costs and health service use were complex to interpret because there was loss to follow up, differing healthcare transfer arrangements and the intervals between the annual visits by the research assistants were not always one year (young people have a lot going on in their lives and often visits had to be rescheduled). Strict econometric modelling, despite considerable efforts, proved intractable to undertake. A less robust exploratory analysis was adopted. This drew out some key implications and generated a balance sheet. This was not wholly in concordance with some conclusions from other parts of the Programme, but triangulation of findings allowed key consistent conclusions and implications to be identified and reported.