Cost-effectiveness analysis of current practice and parent intervention for children under 3 years presenting with expressive language delay

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Record Status
This is a critical abstract of an economic evaluation that meets the criteria for inclusion on NHS EED. Each abstract contains a brief summary of the methods, the results and conclusions followed by a detailed critical assessment on the reliability of the study and the conclusions drawn.

Health technology
The use of a parent-based intervention (PBI) for preschool children with expressive language delay. PBI was defined as an indirect parent-based group treatment that combined the setting of linguistic objectives and an interactive approach. The emphasis of the PBI was the development of the child(s expressive language using daily routines and naturally occurring situations.

Type of intervention
Other: Integrated care.

Economic study type
Cost-effectiveness analysis.

Study population
The study population comprised children with expressive language delay who had been referred to the speech and language department of Portsmouth PCT. The inclusion criteria were:

the child was aged between 22 and 36 months;

the child had little or no expressive language (defined as a vocabulary of 30 or less single words);

there was no formal evidence of verbal comprehension difficulties (more than 2 standard deviations from the norm), or no informal evidence of more than moderate verbal comprehension delay;

the child had not already received speech and language therapy;

the child had been given a diagnosis of either language delay, or speech and language delay; and

the child had no known aetiological factors.

Setting
The setting was a mixture of primary or community, and home-based care. The economic study was carried out in Portsmouth, UK.

Dates to which data relate
The dates to which the effectiveness and resource use related were not stated. The costs were based on 1999 data.

Source of effectiveness data
The effectiveness data were derived from a single study.
Link between effectiveness and cost data
The costing was undertaken prospectively on the same patient sample as that used in the effectiveness study.

Study sample
No power calculations were conducted to determine the sample size. Consecutive patients referred to the department, who met the inclusion criteria, were recruited to the study. There were no refusals to participate and no reported exclusions. A total of 28 children/families started the trial. The first 14 eligible children were offered or allocated to one of two PBI groups. The next 14 children meeting the inclusion criteria were offered or allocated to general care.

Study design
This was a multi-centre, non-randomised controlled trial of 6 months’ duration. Each child was assessed with an 8-month gap between pre- and post-intervention. Allocation to the groups was carried out on the basis that the first 14 referred children who met the inclusion criteria were allocated to the PBI, while the next 14 children were allocated to general care. Parents with children allocated to the PBI were offered the choice of two different PBI groups. Both groups offered the same services, just at different times and locations. Although a different speech and language therapist was used at pre- and post-intervention where possible, blinding was considered implausible because of the general availability of case notes within the clinics and the potential for information transfer from parental involvement at the follow-up visits. Six children/families (21%) did not complete the study. Two of these had been allocated to the PBI (14%) and four to general care (29%).

Analysis of effectiveness
The primary outcome was the measurement of increased linguistic complexity. This was divided into six measures of expressive and receptive language abilities as follows. Parental reports of new vocabulary learnt and length of utterance were used to supplement objective measures. These parental reports included a word list (EST.VOC) and an estimate of phrase length (EST.PL). Parental reports were also used to measure correspondence between home and clinic-based language abilities. Comprehension was measured using the Reynell Developmental Language Scales (RDLS RS). Expression and comprehension were measured using the Pre-School Language Scale-3 UK (PLS C RS for comprehension and PLS E RS for expression). The mean length of utterance (MLU) was derived from a language sample comprising a minimum of 50 utterances from each child.

The basis of the analysis was treatment completers only (22 children). With the exception of the age of the children, the characteristics of the patients (demographic and language abilities) were comparable. Adjustments for pre-intervention differences in child age were made at analysis.

Effectiveness results
The following results are expressed as the mean gain (pre- and post-intervention scores) on the six outcome measures.

The mean gain in estimated vocabulary score was 186.50 (standard error, SE=61.17; 95% confidence interval, CI: 51.87 - 321.13) in the PBI group and 47.60 (SE 17.65; 95% CI: 8.28 - 86.92) in the general care group.

The mean gain in estimated phrase length was 2.82 (SE 0.30; 95% CI: 2.17 - 3.47) in the PBI group and 1.00 (SE 0.21; 95% CI: 0.53 - 1.47) in the general care group.

The mean gain in RDLS RS (comprehension) was 14.36 (SE 2.16; 95% CI: 9.62 - 19.10) in the PBI group and 5.30 (SE 1.26; 95% CI: 2.50 - 8.10) in the general care group.

The mean gain in MLU was 2.01 (SE 0.23; 95% CI: 1.50 - 2.52) in the PBI group and 0.50 (SE 0.18; 95% CI: 0.09 - 0.91) in the general care group.

The mean gain in PLS C RS (comprehension) was 7.36 (SE 1.27; 95% CI: 4.57 - 10.15) in the PBI group and 3.30 (SE
0.73; 95% CI: 1.67 - 4.93) in the general care group.

The mean gain in PLS E RS (expression) was 9.09 (SE 0.86; 95% CI: 7.21 - 10.97) in the PBI group and 4.80 (SE 0.70; 95% CI: 3.25 - 6.35) in the general care group.

Clinical conclusions
Based upon the mean scores gained, both interventions produced improvements in the children's language abilities. However, the results indicated that those who received the PBI made significantly greater language gains than those receiving general care, with the possible exception of estimated vocabulary (see incremental benefits later).

Measure of benefits used in the economic analysis
The measure of benefit used was the cost per language ability score gained.

Direct costs
The health care costs included direct treatment (consultation costs, administration, overheads), parents travel and meeting time, and costs relating to staff. In the PBI group, the costs were estimated on the basis of 8 parents attending. The resource use data were collected from a record sheet given to the therapists. The costs were estimated based upon standard resource cost assumptions from the perspective of the PCT, using cost data obtained from the Financial Directorate of the Trust and 1999 prices. The parents were given a record sheet on which to record travel methods and cost, travel time and meeting time. These data were subsequently pooled (except for the time element), owing to the small sample size. With the exception of the parents' travel costs per mile, it was not possible to separate resource and cost data on any of the items. Discounting was not relevant because of the short-term nature of the study. The study presented both average and incremental costs per language ability score gained. Costs that were identical to the two alternatives (e.g. those relating to pre-and post-assessment meetings) were not included in the analysis (differential costing), while those relating to parents who did not attend sessions were ignored due to their perceived insignificance.

Statistical analysis of costs
The costs were reported as descriptive statistics. For direct treatment costs, these were given as mean values with the SE. For parent costs, these were given as total costs.

Indirect Costs
In line with the perspective chosen, the indirect costs were not included.

Currency
UK pounds sterling.

Sensitivity analysis
No sensitivity analyses were conducted. Although the authors discussed the impact of group size on the costs, and therefore the cost-effectiveness, they highlighted the difficulties in drawing any firm conclusions from this discussion.

Estimated benefits used in the economic analysis
The analysis of differences between the group post-intervention scores showed significant differences in favour of the PBI on all measures, except for estimated vocabulary. The mean gain was: 138.5 (95% CI: -0.27 - 277.98) for estimated vocabulary;

1.82 (95% CI: 1.04 - 2.59) for estimated phrase length;
9.06 (95% CI: 3.7 - 14.42) for RDLS RS;
1.51 (95% CI: 0.88 - 2.14) for MLU;
4.06 (95% CI: 0.92 - 7.2) for PLS C RS; and
4.29 (95% CI: 1.95 - 6.63) for PLS E RS.

Non-parametric testing of all group comparisons produced the same pattern of results. However, the gain in estimated vocabulary was found to be significant at the 5% level.

Cost results
The average total direct treatment cost per child was 96 (SE=2.66) for the PBI and 80.83 (SE=2.05) for general care. The total travel costs to an average parent attending therapy were 31.80 (PBI) and 5.78 (general care), respectively.

Synthesis of costs and benefits
The average total direct treatment cost per child was 96 (SE=2.66) for the PBI and 80.83 (SE=2.05) for general care. The total travel costs to an average parent attending therapy were 31.80 (PBI) and 5.78 (general care), respectively.

The benefits and costs were combined as the costs () per language ability score gained. The results of the incremental analysis ( per additional outcome) were as follows:

for estimated phrase length, 8.77;
for estimated vocabulary, 0.18;
for RDLS RS, 1.60;
for MLU, 10.97;
for PLS C RS, 3.47; and
for PLS E RS, 3.82.

The authors considered the sensitivity analysis to be of little use, given that any changes in key cost parameters will influence the results of both comparators in the same direction.

Authors' conclusions
The authors considered the sensitivity analysis to be of little use, given that any changes in key cost parameters will influence the results of both comparators in the same direction.

CRD COMMENTARY - Selection of comparators
The choice of the comparators was justified on the basis that the potential effectiveness of parent-based language intervention models has been acknowledged in theory and practice, and the general care model represented current practice in the authors' setting. You should decide if these represent widely used technologies in your own setting.

Validity of estimate of measure of effectiveness
The analysis was based on a non-randomised controlled trial. A controlled trial was appropriate given the study question. However, the lack of randomisation and blinding presented significant threats to the internal validity of the study. The authors acknowledged some difficulties in this regard in terms of the nature of the intervention, and some attempts to address the potential sources of bias were made. The lack of power calculations and the subsequent small
sample size meant that it was unclear whether the pre-intervention study sample was sufficient to obtain statistically significant outcome results. Given that consecutive patients were selected, it is likely that the study sample was representative of the targeted study population, although a larger study sample would have given better assurance of this. The groups were reported to be comparable in terms of demographics and language ability, and the numbers were deemed to reflect referral rates in practice.

Loss to follow-up was reported, but the analysis appears to have been conducted on the basis of treatment completers only. This means there is a possibility that the results were overestimated. A further limitation to the strength of inference from the findings was that full compliance with the 11 PBI sessions was not achieved in some cases, whilst in one instance data were not collected at all. Given the limitations highlighted, the internal validity of the study is likely to be poor.

Validity of estimate of measure of benefit
The measure of benefit was a selection of language ability scores measured over an 8-month period. Subjective measures were included to support standardised objective assessments, which were deemed to provide measures of reliability and validity (although no references were given to confirm this).

Validity of estimate of costs
It appears that categories of direct costs relevant to the UK health care perspective were included. In addition, the authors appropriately considered a preliminary analysis of other direct costs accruing to parents in terms of travel and time costs. They also acknowledged that, although outside the scope of this analysis, the adoption of a wider societal perspective (to include, for example, indirect costs for loss of work time) would be a more appropriate strategy. The price year was reported, which will aid any future reflation exercise. The costs and the quantities were not reported separately, with the exception of mileage costs for travel expenses. However, this would not allow the analysis to be easily reworked in other settings. Resource use was obtained from record sheets completed by the therapists and the unit costs were taken from the authors’ setting. No sensitivity analysis of the resources (other than that implicit in group size variation) or statistical analysis of the prices was performed, thus limiting the interpretation of the study findings.

Other issues
The authors compared their findings with those from other studies, which (in terms of effectiveness) were broadly in agreement. However, since no rigorous economic analysis of alternative treatments had been conducted to date, a comparison of the final results was not possible. The issue of generalisability to other settings was not explicitly addressed and no sensitivity analyses were conducted. A justification for the lack of sensitivity analyses was provided. The authors reported a number of limitations to their study. For example, the small sample size and short-term follow up of a condition that might require longer measurement to allow for spontaneous correction.

Implications of the study
The authors suggested that these study findings are of importance to service providers, speech and language therapists, and other professionals working with preschool children with language delay. They suggested that further studies might consider the effects of group size variations upon outcomes as well as costs, in addition to an investigation of how increased benefits might compensate parents for the additional costs of accessing the PBI. The authors stressed the need for a long-term view of preschool language-delay (despite claims that some patients will undergo spontaneous correction without treatment). In this situation, they recommended the inclusion of quality of life indicators in any future analysis of benefits.

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