The outcome of surgical intervention for early deformity in young ambulant children with bilateral spastic cerebral palsy

We reviewed the outcome in 24 children with bilateral spastic cerebral palsy aged seven years or younger for whom surgery was recommended between 1999 and 2005 following gait analysis. A total of 13 children (operative group) had surgery and the remaining 11 (control group) did not, for family or administrative reasons. The operative group had at least two post-operative gait analyses at yearly intervals, with eight children having a third and six children a fourth. The control group had a second analysis after a mean interval of 1.5 years (95% confidence interval 1.1 to 1.9). In the operative group, the Gillette gait index, the ranges of movement in the lower limb joint and knee extension in stance improved following surgery, and this was maintained overall at the second post-operative analysis. The minimum knee flexion in stance in the control group increased between analyses.

These results suggest that surgical intervention in selected children can result in improvements in gait and function in the short to medium term compared with non-operative management.

The management of young ambulant children with bilateral spastic cerebral palsy generally involves the use of physiotherapy, casting and orthoses, with the aim of preventing or deferring the development of deformities in the lower limb. It is considered important to defer surgical intervention to allow maturation of the gait pattern of the child\(^1,2\) and to reduce the risk of a recurrence of deformity with growth.\(^3\)

Although the natural history of mobility in children with bilateral spastic cerebral palsy is one of deterioration,\(^4,5\) children with this condition who walk independently respond well to early non-operative intervention and do not develop deformities until their second decade. They are likely to represent one end of a spectrum, with the other comprising more severely affected ambulant children who develop lower limb deformity early and who may respond less well to conservative management. The different levels of involvement of these groups is reflected by levels in the classification system of gross motor function.\(^6\) This ranges from level I, where mobility is normal, to level V where a child is dependent on a wheelchair for support in sitting. The mobility of a child in a particular level may improve or deteriorate,\(^7\) and this variability affects the formulation of surgical goals.\(^8\) Surgery may be carried out to improve a near normal gait pattern, to maintain or prolong a more limited level of mobility with growth, or to allow a child with bilateral spastic cerebral palsy to develop functional mobility,\(^9\) depending on the child’s level of function and likely prognosis for long-term ambulation. Although the potential role of surgical intervention for children with bilateral spastic cerebral palsy has broadened, evidence to guide the timing, extent and functional goals of surgery in younger ambulant children with this condition is limited. A favourable outcome following multilevel surgery has been described for independent\(^10,11\) and assisted \(^12,13\) walkers but the age ranges of the groups studied are broad, with mean ages of more than eight years, making these findings less applicable to younger children. Similarly, the intensive multilevel surgical procedures which appear to work well in older children, may not be as well tolerated in younger children with limited mobility. Aiona and Sussmann\(^14\) have discussed these factors and suggest that younger children may benefit from staged and initially minor operative procedures. To our knowledge there is no study looking specifically at the outcome of surgery in the younger and more affected group compared with a control group. The ideal approach would be to consider a randomised controlled clinical trial, but this poses ethical difficulties as most young children are referred for consideration of surgical intervention because of a perceived failure.
of non-operative management and deterioration in their level of mobility. Randomising them into what may, in the absence of an alternative, be seen as treatment and non-treatment groups, may not be accepted by the families or the referring clinicians. A randomised controlled trial may be more acceptable for older and more functional children, but the functional goals and outcome of surgery in this group may be different from those in younger and more affected patients.

Surgical intervention in older ambulant children with bilateral spastic cerebral palsy has been shown to maintain or improve function compared with conservative management, with a deterioration in function being noted in children for whom operation was recommended but not performed. We hypothesised that younger children, aged eight years or less, with bilateral spastic cerebral palsy who had surgery, would also show an improvement in gait which would be maintained for at least two years following operation, and that children in whom this was recommended but not performed would show a deterioration in mobility.

**Patients and Methods**

Our unit acts as a specialist assessment and treatment centre for children with cerebral palsy, providing gait analysis, conservative management in close association with the community teams, and operative intervention. Children are referred for gait analysis in order to guide management, and are followed up with repeat analyses. We reviewed all the ambulant children aged seven years or younger at presentation with bilateral spastic cerebral palsy in whom multilevel surgical intervention had been recommended following gait analysis between 1999 and 2005. Children for whom surgical intervention was not thought to be needed, or those in whom it was thought that it would be unlikely to provide a benefit because of the extent of their involvement, were excluded. We identified 14 children in whom surgery had been performed and who had at least two gait analyses at yearly intervals following operation (operative group). One child was excluded because of missing anthropometric data from the first (pre-operative) analysis. However, his post-operative outcome was similar to that of the other children in this group. We assessed 11 children in whom surgery had been recommended but not performed (control group). In seven of these the initial referral was by a community team, and was followed by discussion within the team and family before a subsequent referral for orthopaedic review. In two cases a continuation of conservative treatment was preferred, and in the remaining two the family did not wish to consider surgical intervention. All of the children in the control group had a follow-up gait analysis at a mean of 1.5 years later (95% CI 1.1 to 1.9). The children were evenly distributed throughout the region, with most community teams being involved in the care of children from both groups.

Each gait analysis was performed by the same team according to a standardised protocol which included a clinical history and examination followed by the acquisition of video, three-dimensional movement, electromyographic and kinematic data (for independent walkers) using a Vicon 612 optical motion capture system (Vicon, Oxford, United Kingdom). A minimum of four walking trials were recorded and assessed, with the children walking barefoot at a self-selected speed and using assistive devices as needed. The recommendations for treatment were made by the same orthopaedic surgeon (MG) and the operations were carried out in our unit. The most frequent procedure recommended and performed was bilateral lengthening of the hamstrings and calf muscles (Table I). In the first year of the study, children used ankle-foot orthoses after a six-week period in below-knee casts, and they had knee extension splints as needed for the first three to six months after surgery. In the following years, knee extension splints were not used after surgery, and the children were encouraged to walk in shoes after four weeks in a cast. The post-operative management was otherwise similar, with weight-bearing beginning on the second post-operative day, with close liaison between the physiotherapists in the hospital and the community. The data reviewed for each child included the level of mobility, details of previous non-operative management, the popliteal angles, the maximum degree of dorsiflexion available with the knee in extension, and the operative procedures recommended. Because of the interdependency between the joints of the lower limbs in gait, individual joint kinematics were not analysed, with the exception of minimum knee flexion in stance. Instead, the Gillette gait index was calculated for each child. This index uses multivariate analysis to derive a single figure from 16 variables, including movement data and normalised velocity, with a greater value being interpreted as a greater variation from normal gait. The Gillette gait index has been used to describe the outcome of multilevel surgery in children with diplegia, with an improvement or deterioration of more than 10% of the pre-operative value following intervention considered as significant. The index appears to be reproducible between individual gait laboratories. The typically-developing reference group in our laboratory had a mean Gillette gait index of 65.9 (95% confidence interval (CI) 35.2 to 96.7). The movement data were reviewed and the Gillette index values calculated by the second author (PS), who was not involved in the acquisition of data or in the clinical management of the children in the study. We reviewed the minimum knee flexion in stance, as this is not specifically included in the Gillette gait index and appears to significantly influence the support moment in the lower limbs.

The study was approved by the hospital research ethics committee and used retrospective anonymised data which had been gathered as part of routine clinical management.

Statistical analysis was performed using GraphPad Prism 4.0 for Macintosh (GraphPad Software Inc., San Diego, California). The Shapiro-Wilks test was used to assess whether individual datasets were consistent with a normal
distribution: groups were then compared using an unpaired t-test or Mann-Whitney U test as appropriate. Paired data were assessed using the paired t-test or Wilcoxon's matched-pairs signed ranks test, and sequential data were assessed using a repeated measures analysis of variance (ANOVA) or the Friedman test, with Tukey and Dunn's post hoc analysis, respectively. Because of the limitations of retrospective power calculations, the effect sizes were calculated with Hedges' correction for small sample size.$^{19}$ A p-value < 0.05 was considered statistically significant.

Results

All the children in the operative group and seven in the control group were male. Nine had botulinum toxin A injections prior to gait analysis; five had injections only, four had injections and serial casting, and two had casting only. In the control group, two children had serial casting, five had botulinum toxin A injections, and three had casting and botulinum toxin A injections prior to referral. One child in the control group did not have casting or injections prior to the first analysis. The mean interval between the first gait analysis and surgery in the operative group was 0.7 years (95% CI 0.5 to 0.9), and the mean interval between surgery and the first post-operative analysis was 1.2 years (95% CI 1.1 to 1.3), with subsequent post-operative analyses being performed approximately yearly.

The operative and control groups are compared in Table II. The children in the control group had less marked equinus deformities and a greater degree of knee extension in stance than those in the operative group at the first analysis. The initial Gillette gait index grades were similar in both groups.

Isolated soft-tissue surgery was recommended initially in 12 of 13 patients in the operative group because of concerns about their ability to rehabilitate following surgery. Two of the children in this group who had previously been considered for bilateral calcaneal osteotomy, had these procedures performed respectively 1.5 years and 2.5 years after a successful outcome from the initial surgery. The children in the control group had ongoing physiotherapy, and a number of them had increased physiotherapy support, including an exercise programme between analyses. During the interval between gait analyses, two children in the control group had botulinum toxin A injections, one for the first time.

The results from the first post-operative analysis in the operative group and those from the second analysis in the control group are compared in Table II. The operative group showed improvements in passive ranges, including the maximum degree of dorsiflexion and the popliteal angle, in minimum knee flexion in stance, and in their Gillette gait index values. The control group showed a deterioration in minimum knee flexion in stance between analyses. All 13 children in the operative group had pre-operative and two post-operative analyses. The parameters assessed all improved after surgery and remained so at the second post-operative gait analysis in the case of Gillette gait index (Friedman test, $p = 0.001$), minimum knee flexion in stance (repeated measures ANOVA, $p < 0.001$) and maximum degree of dorsiflexion (Friedman test, $p < 0.0001$). The popliteal angle improved initially after surgery, but was similar to the pre-operative level at the second post-operative analysis. Individual changes in Gillette gait index within the two groups are shown in Figure 1.

The data from the third (n = 8) and fourth (n = 6) post-operative analyses have not been analysed statistically because of the limited numbers involved, but the results are

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**Table I. Details of the operative group**

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age at presentation (yrs)</th>
<th>GMFCS level</th>
<th>Age at operation (yrs)</th>
<th>Procedure</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>5</td>
<td>II</td>
<td>5.9</td>
<td>HBL, BGS</td>
<td>Repeat HBL, BGS following two-year post-operative analysis</td>
</tr>
<tr>
<td>2</td>
<td>6.5</td>
<td>III</td>
<td>7.8</td>
<td>BPL, HBL, BGS</td>
<td>Completed two-year post-operative analysis</td>
</tr>
<tr>
<td>3</td>
<td>7.5</td>
<td>IV</td>
<td>8.2</td>
<td>RPL, HBL, RGS</td>
<td>BTAL following two-year post-operative analysis</td>
</tr>
<tr>
<td>4</td>
<td>6.9</td>
<td>II</td>
<td>7.7</td>
<td>HBL, BGS</td>
<td>Completed two-year post-operative analysis</td>
</tr>
<tr>
<td>5</td>
<td>6.2</td>
<td>III</td>
<td>6.9</td>
<td>BBL, BGS</td>
<td>Completed two-year post-operative analysis</td>
</tr>
<tr>
<td>6</td>
<td>7.1</td>
<td>III</td>
<td>8.2</td>
<td>BPL, BBL, RGS</td>
<td>Completed two-year post-operative analysis</td>
</tr>
<tr>
<td>7</td>
<td>5.8</td>
<td>II</td>
<td>7.2</td>
<td>HBL, BGS</td>
<td>Repeat HBL and BTAL following three-year post-operative analysis</td>
</tr>
<tr>
<td>8</td>
<td>6.9</td>
<td>II</td>
<td>7.8</td>
<td>BBL, BGS</td>
<td>Completed four-year post-operative analysis</td>
</tr>
<tr>
<td>9</td>
<td>5.2</td>
<td>II</td>
<td>5.7</td>
<td>BHL, LCO, LGS</td>
<td>Completed four-year post-operative analysis</td>
</tr>
<tr>
<td>10</td>
<td>6.8</td>
<td>III</td>
<td>6.9</td>
<td>BPL, BBL, BHS</td>
<td>Repeat HBL and RTAL following four-year post-operative analysis</td>
</tr>
<tr>
<td>11</td>
<td>7.7</td>
<td>II</td>
<td>8.1</td>
<td>BGS</td>
<td>Completed five-year post-operative analysis</td>
</tr>
<tr>
<td>12</td>
<td>5.5</td>
<td>II</td>
<td>5.5</td>
<td>RPL, HBL, BGS</td>
<td>Completed five-year post-operative analysis</td>
</tr>
<tr>
<td>13</td>
<td>6.25</td>
<td>III</td>
<td>6.6</td>
<td>HBL, BRFT, BGS</td>
<td>Repeat HBL following five-year post-operative analysis</td>
</tr>
</tbody>
</table>

* GMFCS, gross motor functioning classification system
† BHL, bilateral distal hamstring lengthening; BGS, bilateral gastrocnemius slides; BPL, bilateral psoas lengthening; RPL, right psoas lengthening; LCO, left calcaneal osteotomy; LGS, left gastrocnemius slide; BRFT, bilateral rectus femoris transfer
‡ BTAL, bilateral tendon Achilles lengthening; RTAL, right tendon Achilles lengthening
shown in Table III and suggest that the improvements gained have been maintained overall, although, as shown in Table I, four children have needed repeat surgery, one after the second post-operative analysis, one after the third, one after the fourth and one after the fifth.

Although the gross motor function classification system is a prognostic tool rather than an outcome measure, it was noted that at their last analysis three children in the operative group had improved their motor classification level by one grade, and the remainder had a similar grade to their pre-operative assessment. At their second analysis, eight children in the control group had a similar gross motor function classification system level, but three had deteriorated by one grade. On assessment of outcome as defined by the Gillette gait index, 12 of the children in the operative group had an improvement of more than 10% compared with their pre-operative index; the other child had a deterioration of more than 10%, but this was the only child in the study with a pre-operative gross motor function classification system level of IV. In the control group, four children improved their Gillette gait index by more than 10%, one remained stable, and six showed a deterioration between analyses of more than 10% of their pre-operative Gillette gait index. The control group also had a significant increase in minimum knee flexion in stance between analyses (t-test, p = 0.045).

**Discussion**

The results support our hypothesis that limited surgical intervention in ambulant children with bilateral spastic cerebral palsy presenting at the age of seven years or younger, with multilevel deformities of the lower limb
which have not responded to non-operative management, can stabilise or prolong mobility. We do not suggest that early surgical intervention should be routinely performed or be considered as an alternative to accepted non-operative management when this appears to be effective, but consider that limited surgery with conservative goals should be considered in this group if their methods of treatment do not appear to be effective.

There are, however, a number of limitations to this study. It describes a very specific group of children with bilateral spastic cerebral palsy treated with relatively limited surgery in a specialist centre, and the conclusions may not be generalised to other age groups or other surgical approaches. The operative group had greater knee flexion in stance and a greater degree of fixed equinus, suggesting greater involvement, and had a longer interval between their first and second analyses. This suggests that the children in this group might have been more likely to have a deterioration in function compared with those in the control group which would have led to an underestimation rather than an overestimation of the effect of surgery. The group sizes are small, and the length of follow-up of the patients in the operative group was limited, with some children completing only two post-operative analyses. The changes seen in the gross motor function classification system between analyses may relate to the heterogeneity of this group of children: variations in the functional level of a child and in their level of mobility within a particular level have been reported in longitudinal studies. The changes noted in the level do appear to reflect the changes seen in the Gillette gait index.

The variability of the response of individual muscle groups to surgical intervention is interesting. Although lengthening of the calf muscles was aimed at undercorrection rather than overcorrection, which may explain the number of children who needed repeat surgery to these muscles, the surgery had a longer-lasting effect on the passive length of the calf muscles than did comparable surgery on the hamstrings. The hamstring length in the operative group was improved at the first post-operative analysis, but had returned to its pre-operative length by the second assessment. This did not appear to be associated with a reduction in knee extension in stance between the first and second analyses, but may be related to the long-term development of a crouch-gait with growth, although the available data for up to four years following surgery do not suggest an increase in knee flexion over this period. The variation in the individual Gillette gait index values seen in

**Table III. Follow-up data for the operative group (mean, 95% confidence interval)**

<table>
<thead>
<tr>
<th>Post-operative interval</th>
<th>2 years (n = 13)</th>
<th>3 years (n = 8)</th>
<th>4 years (n = 6)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gillette gait index</td>
<td>794 (354 to 1233)</td>
<td>648 (289 to 1006)</td>
<td>652 (291 to 1013)</td>
</tr>
<tr>
<td>Minimum knee flexion in stance (°)</td>
<td>22 (19 to 27)</td>
<td>20 (16 to 25)</td>
<td>23 (19 to 27)</td>
</tr>
<tr>
<td>Popliteal angle (°)</td>
<td>67 (63 to 70)</td>
<td>68 (62 to 73)</td>
<td>67 (61 to 73)</td>
</tr>
<tr>
<td>Maximum passive dorsiflexion (°) (knee extended)</td>
<td>2 (2 pf to 5 df)</td>
<td>1 (3 pf to 5 df)</td>
<td>5 (0 to 10)</td>
</tr>
</tbody>
</table>

* pf, plantar flexion; df, dorsiflexion
the control group between analyses and in the operative group following surgery, is also interesting. A smaller (i.e. ‘superior’) Gillette gait index pre-operatively did not necessarily imply a better outcome post-operatively. The rehabilitation and community support following surgery were similar for all children, and factors such as muscle strength and selective muscle control\textsuperscript{7,21} may be important in predicting outcome. Other factors, such as the motivation of each child and their ability to participate in the necessary post-operative rehabilitation programme, are likely to be important,\textsuperscript{22} although these may be more difficult to quantify. The child in the operative group who had a deterioration in his Gillette gait index was the only one in either group with an initial gross motor functional classification system level of IV, suggesting that the functional goal of surgery for children with this level of involvement needs to be limited, although an improvement in mobility with this severity of complaint can be achieved following surgery.\textsuperscript{9,13}

We did not assess the effect of surgery on the functional abilities and health-related quality of life for each child, and future prospective studies will be needed for this. The early development of lower limb deformities that have not responded to non-operative management raises concern about the long-term prognosis for mobility of these children: it will be important to follow this cohort to define the duration of effect of surgery and to help define the factors that predict how individual children will respond to operation. The goals and outcome of surgery were limited but are still valid. Although employment and independence in adults with cerebral palsy appear to be influenced by factors other than their level of mobility,\textsuperscript{23,24} maintenance of the ability to walk for short distances indoors, to transfer into and out of a wheelchair, and to stand for toileting at skeletal maturity are likely to enhance their functional independence.

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

References