Introduction

Spina bifida myelomeningocele, a congenital condition characterised by cystic protrusion of the spinal cord through unfused vertebral arches, has been recognised as the second most physically handicapping condition of childhood after cerebral palsy (Anderson 1976). In the past, upper limb function in these children was assumed to be normal, since most of the myelomeningocele lesions as such do not interfere with upper extremity function (Henderson 1968). However, recent studies in the area of hand function and handwriting (Minns et al 1977, Pearson et al 1988, Sand et al 1974) have highlighted the difficulties these children experience with activities of daily living. For example, using the Developmental Hand Function Test, Grimm (1976) discovered that 82% of children with spina bifida scored two standard deviations below control children. Furthermore, children with myelomeningocele were shown to be slower than the controls ($F_{1,280} = 15.49$, $p < 0.001$). The impaired kinaesthetic awareness found in this study is considered to be one of the factors behind the poor hand function observed in children with myelomeningocele. [Hwang SJ, Kentish M and Burns Y (2002): Hand positioning sense in children with spina bifida myelomeningocele. Australian Journal of Physiotherapy 48: 17-22]

Key words: Hand; Kinesthesis; Meningomyelocele; Proprioception

Hand positioning sense in children with spina bifida myelomeningocele

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This study was undertaken to establish whether children with myelomeningocele have abnormal kinaesthesia of the hands. Twenty-one children with myelomeningocele and 21 control children, aged between six and 12 years, were involved in the study. The level of kinaesthetic awareness in the hands was measured by examining the child's ability to copy hand positions, using visual cueing and kinaesthetic cueing. Both accuracy and speed of copying hand gestures were assessed. Children with spina bifida were significantly less accurate in achieving hand positions than the control group ($\chi^2_{10} = 22.60$, $p < 0.001$), with 73% of the children with spina bifida achieving accurate replications compared with 87% in the control group. Furthermore, children with myelomeningocele were shown to be slower than the controls ($F_{1,280} = 15.49$, $p < 0.001$). The impaired kinaesthetic awareness found in this study is considered to be one of the factors behind the poor hand function observed in children with myelomeningocele. [Hwang SJ, Kentish M and Burns Y (2002): Hand positioning sense in children with spina bifida myelomeningocele. Australian Journal of Physiotherapy 48: 17-22]

Kinaesthesia has been suggested to play a role in the acquisition and performance of skilled hand movements, including handwriting (Lynch et al 1992). In handwriting, kinaesthetic feedback is thought to reduce the child's reliance on visual monitoring of pencil control, and thus enable the child to direct visual attention to letter formation, letter order and sentence construction (Benbow 1995). This implies that the child does not need to concentrate on the position of the hand but instead, can concentrate on planning the movement and making any necessary adjustments, which subsequently enhances the speed and accuracy of handwriting (Hamilton 1991). This view is supported by Bairstow and Laszlo (1981), who found a strong positive correlation between kinaesthetic ability, and drawing and writing in young school children. Furthermore, these authors proposed that the handwriting difficulties experienced by some children could be partly attributed to an inadequate ability to process kinaesthetic information (Bairstow and Laszlo 1981).

As kinaesthetic awareness in the hands is important for fine motor skills, it is conceivable that impaired kinaesthetic feedback can have detrimental and wide-ranging effects on the developing child, particularly on the functional use of the hand. Determining whether abnormal kinaesthesia of the hands is present in children with myelomeningocele may shed further light on why these children experience difficulties with hand function. Thus the purpose of the current study was to compare the performance on copying hand positions between a group of children with myelomeningocele and a group of normal children. It was hoped that this information would contribute to the advancement of knowledge in the areas of assessing and treating children with spina bifida.
Methods

Subjects  This study involved two subject groups. The experimental group consisted of 21 children with myelomeningocele, aged between six and 12 years (mean age = 9.1, SD = 1.8). The 12 females and nine males in the experimental group were recruited from Queensland’s Statewide Paediatric Rehabilitation Service and the Queensland Association of People with Spina Bifida or Hydrocephalus. The control group consisted of 21 normal children who had been individually matched with children in the experimental group on the basis of age and gender. Inclusion in the experimental group required subjects to have written informed parental consent, shunted hydrocephalus and lesions below the level of T4. In addition, a screening test, which involved repetitive and successive finger-thumb opposition, was administered to ensure that these children had isolated finger movements for fine motor control. Children from both the experimental and the control group were excluded from the study if they had visual, auditory or intellectual impairment. Subjects with other neurological deficits, concomitant musculoskeletal problems, or major injury to the upper limbs within the last year were also excluded from the study. Ethical approval for the study was received from the Medical Ethics Committees of The University of Queensland and the Royal Children’s Hospital, Brisbane.

Procedure  The test was conducted in a quiet and well-lit room, with the subject comfortably seated using a table and chair of appropriate height. A masking box was placed on the table to occlude the subject’s vision, while allowing the examiner to have a full view of the subject’s hands. The box had an internal shelf, which enabled the forearms to be supported along predetermined lines drawn at an angle of 20 degrees to the table edge, and the hands were hanging freely over the edge of the shelf. Hand preference was ascertained according to which hand the child preferred to use to perform at least three of the five activities described by Denckla (1973). The preferred hand was then recorded and tested for kinaesthesia of the hand.

The level of kinaesthetic awareness in the hands was measured by the subject’s ability to copy hand positions. This was examined by looking at two different methods to cue the child to reproduce hand gestures, these being visual and kinaesthetic cueing. The examiner demonstrated a set of eight visually-cued hand positions, and the child was instructed to produce mirror images of these gestures with the preferred hand, which was hidden inside the masking box. The same patterns were then presented kinaesthetically, but this time the masking box concealed both the test and non-test hands. In the kinaesthetically-cued situation, the examiner positioned the non-test hand, and requested the child to duplicate the pattern with the preferred hand. Each subject was instructed to produce each hand position only once under each cueing condition, and to copy the hand position as quickly and as accurately as possible. Before the testing began, a practice position was given to ensure the subject’s understanding of the task.

The order of the two sets of cues and the eight testing patterns within each set were randomised. The eight testing positions were based on those described by Lynch et al (1992), and later adapted by Grant and Watter (1998). These positions are presented in Figure 1.

The time taken to copy each test pattern was recorded in seconds rounded to two decimal places, and the accuracy of replication was graded using the criteria described by Lynch et al (1992), where:

0 = failure to move the hand from the resting position.
1 = no resemblance to the test position.
2 = incomplete replication, which may include use of the wrong fingers in the correct relationship, one finger out of place, inappropriate opposition, or a reversal of gesture.
3 = complete and accurate replication.

Any position which could not be held by the non-test hand during kinaesthetic cueing was also noted.

A major limitation with existing tests of kinaesthetic awareness has been the failure to incorporate both qualitative and quantitative measures. This problem was apparent in a study by Berges and Lezine (1965), where the authors failed to take into account the time taken to copy hand gestures. It is conceivable that it could have been

![Figure 1. Hand positions.](image-url)
time-consuming for the child to copy the position, but a relatively high score could still occur if the child had accurately imitated the movement. Consequently the measurement utilised in this experiment had incorporated both qualitative as well as timed measures. Furthermore, the test used in this experiment eliminated short-term memory as a confounding variable, by having the cues present throughout the time required to replicate the hand positions.

**Statistical analysis** Data analysis was performed using Minitab 11 statistical package. Ordinal logistic regression (Armstrong and Sloan 1989) was chosen to examine the effects of the independent variables on the accuracy scores, using two levels of children groups, two levels of cues, and eight levels of hand positions. Results were considered statistically significant when \( p \) values were less than 0.05. To permit the use of parametric statistics, the time scores were transformed by taking natural logarithm of the time scores. The effect of group, cue and hand position on the time taken to assume the test positions were examined using repeated measures ANOVA.

**Results**

**Reliability study** To establish test-retest reliability, the procedure was administered to a group of 10 normal children aged from six to 12 years, on two occasions, separated by an interval of one week. The results were then analysed by comparing the time scores obtained on the two occasions using ANOVA. The time scores on the two trials did differ significantly \((F(1,144) = 8.18, p = 0.005)\), with a mean in logarithm seconds (SD) of 1.07 (0.54) on the first trial, versus 0.89 (0.55) on the second trial. This improvement may represent a learning effect of the first on the second test. Even though there was a difference between the two trials, both the cues \((F(1,144) = 0.04, p = 0.833)\) and the hand positions \((F(7,144) = 1.79, p = 0.093)\) had no significant effect on these differences. Reliability of the test was also supported by intraclass correlation coefficient (2,1) of 0.65 (95% CI = 0.45 to 0.77) for cues that were presented visually and 0.72 (95% CI = 0.57 to 0.82) for cues that were provided kinaesthetically.

**Performance grades** Ordinal logistic regression was performed on the accuracy scores as described earlier. The results revealed that there were significant differences between the two groups of children \((\chi^2(1) = 22.60, p < 0.001)\), the two types of cues \((\chi^2(4) = 8.26, p = 0.004)\) and the eight hand positions \((\chi^2(7) = 144.28, p < 0.001)\).

When overall scores were considered, children with myelomeningocele were significantly less accurate than children in the control group, with 73% of the children with spina bifida achieving a Grade 3 accuracy score, compared with 87% in the control group. The percentage of subjects in each group who achieved a complete and accurate replication for each test position (ie a Grade 3 accuracy score) is shown in Table 1. When the cues were considered separately, children with spina bifida still performed less accurately than the control group on visually-cued positions. Similarly, there is an even smaller proportion of children with spina bifida achieving a complete and accurate replication than the control children, when the cue was provided kinaesthetically. It is also apparent from Table 1 that for 11 of the 16 position and cue combinations, children with spina bifida performed less accurately than the control group.

When overall scores were considered, children with myelomeningocele revealed significant differences between the two groups of children \((F(1,280) = 15.49, p < 0.001)\), the cues \((F(4,280) = 17.28, p < 0.001)\) and the hand positions \((F(7,280) = 37.27, p < 0.001)\). There appeared to be a small but significant interaction effect between group and position \((F(14,280) = 2.53, p = 0.015)\), but no significant interaction was observed between group and cue \((F(1,280) = 0.83, p = 0.369)\).

Overall, children with spina bifida were shown to be significantly slower at reproducing hand gestures than were the control children, with the means in logarithm seconds (SD) being 1.42 (0.78) and 1.00 (0.58) respectively. Group means and standard deviations of the time scores are summarised in Table 2. When the cues were examined separately, children in the experimental group were still less proficient than the control group at copying hand positions that were provided kinaesthetically. This difference was even more pronounced in the visually-cued positions. As illustrated in Table 2, it is interesting to note

### Table 1. Percentage of subjects attaining Grade 3 accuracy score for each test position.

<table>
<thead>
<tr>
<th>Cues</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>Average</th>
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<tr>
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<tr>
<td>Spina bifida</td>
<td>57</td>
<td>100</td>
<td>62</td>
<td>91</td>
<td>62</td>
<td>67</td>
<td>95</td>
<td>100</td>
<td>79</td>
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<tr>
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<td>81</td>
<td>100</td>
<td>81</td>
<td>91</td>
<td>76</td>
<td>81</td>
<td>95</td>
<td>100</td>
<td>88</td>
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<tr>
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<td></td>
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<td></td>
</tr>
<tr>
<td>Spina bifida</td>
<td>48</td>
<td>91</td>
<td>43</td>
<td>71</td>
<td>24</td>
<td>71</td>
<td>81</td>
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<td>Controls</td>
<td>76</td>
<td>100</td>
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<td>81</td>
<td>62</td>
<td>91</td>
<td>100</td>
<td>100</td>
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that children with spina bifida were consistently slower than the control group for 15 of the 16 position and cue combinations.

Discussion

The results obtained from this study support the experimental hypothesis that children with myelomeningocele show difficulties with kinaesthesia of the hands, compared with their equivalent norms of the same age and gender. A smaller proportion of children with spina bifida were able to accurately replicate the hand positions. Furthermore, it was also observed that children with myelomeningocele took significantly longer to assume hand positions than the control group.

The results of the current study accord well with a study by Hamilton (1991) who examined sensory hand function in 22 children with myelomeningocele aged between six and 17 years. It was found that more than 40% of these children were unable to detect small joint displacements. It was also documented that the sensory test that presented the greatest difficulty to children with myelomeningocele was that of position sense, with 50% of the children having scores indicative of impairment. However, in Hamilton’s study (1991), the subject’s hand was placed in a particular position, disarranged, and then the child was requested to resume the original position. This implies that the subject’s short-term memory was not eliminated as a possible confounding variable.

Within the present study, it is difficult to identify which of several possible factors account for the high incidence of abnormal kinaesthesia observed in children with spina bifida. A number of factors which have been described in the past as possible causes of hand problems in children with spina bifida include hydrocephalus (Wallace 1973), cerebellar abnormalities associated with the Arnold-Chiari malformation (Anderson 1976), level of the spinal lesion (Grimm 1976), intellectual impairment (Sand et al 1974), visuoperceptual difficulties (Brunt 1980), muscle weakness (Muen and Bannister 1997) and sensory dysfunction (Hamilton 1991).

Poor kinaesthesia of the hands may be seen as either a direct or indirect result of hydrocephalus. Hydrocephalus and the subsequent raised intracranial pressure have been reported to cause stretching of the motor and sensory fibres as they pass around the distended ventricles, as well as causing thinning of the cerebral cortex (Welch and Lorenzo 1991). It is conceivable that such a mechanism of damage to the sensory nerve fibres may contribute to the poor kinaesthetic acuity of the hands in children with spina bifida and hydrocephalus. However, this view is not supported by Muen and Bannister (1997) who argued that hydrocephalus cannot be postulated as the sole factor in causing neurological abnormality of the hands, since children with isolated hydrocephalus did not score significantly differently from the control group on proprioception.

Another CNS anomaly that may explain poor performance on copying hand positions is the Arnold-Chiari malformation. The essential features of this condition include caudal displacement of the medulla, the inferior aspect of the cerebellum, the pons and the fourth ventricle into the upper cervical spine canal (Gilbert et al 1986). It has been reported that cranial and cervical nerve roots may be compressed and exit below their respective foramina in patients with the Arnold-Chiari malformation (Mackenzie and Emery 1971). These anomalies can lead to progressive

<table>
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<th>Cues</th>
<th>1</th>
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<tr>
<td>Spina bifida</td>
<td>1.83</td>
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<td>1.49</td>
<td>2.11</td>
<td>1.79</td>
<td>1.21</td>
<td>0.87</td>
<td>1.55</td>
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<td>Controls</td>
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<td>1.21</td>
<td>0.97</td>
<td>1.43</td>
<td>1.20</td>
<td>0.96</td>
<td>0.70</td>
<td>1.08</td>
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<tr>
<td>Kinaesthetic</td>
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<tr>
<td>Spina bifida</td>
<td>1.73</td>
<td>0.72</td>
<td>1.33</td>
<td>1.59</td>
<td>1.79</td>
<td>1.50</td>
<td>1.09</td>
<td>0.51</td>
<td>1.28</td>
</tr>
<tr>
<td>Controls</td>
<td>1.23</td>
<td>0.83</td>
<td>0.85</td>
<td>1.10</td>
<td>1.30</td>
<td>0.96</td>
<td>0.65</td>
<td>0.36</td>
<td>0.91</td>
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</table>
narrower age range will increase the strength of the results. However, there were a number of changes that could be addressed in future research. The testing procedure used in the present study was considered to have adverse impacts on hand function. Further research, which determines the relationship between kinaesthetic sensitivity of the hands and the conclusions which can be drawn. It should also be noted that this method of assessing hand kinaesthesia is dependent on many closely interwoven factors. Therefore, a deficit in one of these factors, such as muscle weakness and visuoperceptual difficulties, may lower the test scores and obscure the actual level of kinaesthetic functioning in the child. It is only by considering the child’s performance on copying hand positions in the context of an overall neurodevelopmental assessment that firm conclusions can be drawn regarding the kinaesthetic functioning of the child.

In addition, it appears that the impaired kinaesthetic feedback found in this study may be one of the factors underlying the poor hand function observed in children with myelomeningocele. Because of the possible dorsal tract involvement with cord cavitation, this may also account for the impaired kinaesthetic sensitivity of the hands observed in the majority of children with myelomeningocele studied.

It has been argued that kinaesthesia of the hands plays a role in the skill acquisition and performance of fine motor activities, such as dressing, eating and handwriting (Hamilton 1991). As kinaesthesia is important for fine motor skills, it is conceivable that impaired kinaesthetic feedback may have an adverse impact on hand function, limiting the process by which movement patterns are refined and co-ordinated (Laszlo and Bairstow 1985). For example, Bairstow and Laszlo (1981) suggested that the handwriting difficulties seen in some children could be explained by an inadequate ability to process kinaesthetic information. This view is shared by Ziviani et al (1990), who discovered that handwriting speed, alignment and letter formation were the features most detrimentally affected in children with spina bifida and these last two features were significantly influenced by kinaesthetic sensitivity. Therefore, it appears that the deficient kinaesthetic awareness reported in the present study may be one of the factors behind the poor handwriting observed in children with myelomeningocele.

Similarly, other activities involving the functional use of the hand such as doing up buttons, wheelchair propulsions and feeding may also be affected by deficits in kinaesthetic perception. Difficulties in any of these activities may lead to frustration and lower self-esteem in the child (Benbow 1995).

The testing procedure used in the present study was reliable, relatively quick and easy to administer and required minimal equipment. However, there were a number of changes that could be addressed in future investigations. For example, a larger sample size with a narrower age range will increase the strength of the results and the conclusions which can be drawn. It should also be noted that this method of assessing hand kinaesthesia is dependent on many closely interwoven factors. Therefore, a deficit in one of these factors, such as muscle weakness and visuoperceptual difficulties, may lower the test scores and obscure the actual level of kinaesthetic functioning in the child. It is only by considering the child’s performance on copying hand positions in the context of an overall neurodevelopmental assessment that firm conclusions can be drawn regarding the kinaesthetic functioning of the child.

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