Sprengel's shoulder treated by the Woodward procedure: analysis of factors affecting functional and cosmetic outcome

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Abstract

Purpose To report the functional and cosmetic results of cases with Sprengel's shoulder who underwent the Woodward procedure.

Materials and methods Twelve children were operated at a mean age of 5.58 years and reviewed at an average follow up of 31.83 months.

Results The mean preoperative Cavendish grade for cosmetic evaluation was 3.17, which decreased to 1.25 postoperatively (statistically significant, $p < 0.0005$, Wilcoxon signed-rank test). Similarly, the range of abduction at the shoulder increased from a mean of 115.83° preoperatively to 153.33° at final review ($p < 0.0005$). Age had a negative correlation with both functional improvement ($r = -0.55$, Spearman correlation coefficient) and cosmetic improvement ($r = -0.11$), although the latter suggested a very weak association, if any. Cavendish grade improvement and increase in abduction had a strong positive association ($r = 0.713$). However, there was no correlation between the increase in abduction and lowering of the scapula achieved ($r = 0.131$). The presence of an omovertebral bar did not affect the final functional outcome, nor did the presence of associated congenital anomalies.

Conclusion The Woodward procedure is a reliable method for obtaining uniformly predictable results in

Sprengel's shoulder. The surgery should be performed at a younger age in order to optimise the functional outcome.

Keywords Sprengel's shoulder · Woodward procedure · Scapula · Congenital

Introduction

Sprengel's shoulder or congenital elevation of the scapula results from the failure of descent of the scapula during foetal life. The exact cause of interruption of caudad migration of the developing scapula remains obscure [1]. Various degrees of scapular hypoplasia also accompany the process. A large range of congenital malformations may accompany congenitally elevated scapula, such as Klippel–Feil syndrome, rib anomalies, hemivertebrae and scoliosis [2–4].

The deformity which is usually obvious at birth progresses as the child grows. The concern regarding the disorder is chiefly cosmetic and restriction of abduction at the shoulder joint. As a result of rotation of the distal angle of the scapula medially, the glenoid faces inferiorly and, subsequently, the range of abduction at the shoulder is reduced. Binding of the scapula to cervical vertebrae as a result of an omovertebral process reduces the scapulothoracic movement, further impeding the arc of abduction at the shoulder joint [5].

A plethora of surgeries aimed at correction of the deformity have been described. Putti [6] proposed excision of the protruding part of the scapula and lowering of the rest by dividing muscle attachments. This procedure was modified by Green [7] and Woodward [8]. Mears [9] described a surgical technique which involved scapular osteotomy, partial excision of the scapula and release of the

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Fig. 1 Sketches showing the various Cavendish grades. **a** Cavendish grade 1, where the shoulders are level and deformity is not noticeable with the patient dressed. **b** Cavendish grade 2, where the shoulders are almost level, but the deformity is visible with the patient dressed.

c Cavendish grade 3, where deformity is easily visible and the involved shoulder joint is elevated by 2–5 cm. **d** Cavendish grade 4, where the superior angle of the scapula is close to the occiput

long head of triceps. The relative merits and demerits of each procedure exist with proponents of each, and since the deformity is quite rare, there has been no consensus on the optimal management method. This retrospective study is aimed at the evaluation of cosmetic and functional results in children of Sprengel's shoulder treated by the Woodward procedure.

Materials and methods

This series included children with congenitally elevated scapula who underwent the Woodward procedure. All the surgeries were performed by the senior author (P.G.). A total of 12 patients, all with unilateral deformity, were operated between September 2006 and September 2011. The children were evaluated for deformity using the Cavendish grade [10] as a measure of cosmesis and range of abduction at the shoulder as a measure of function. Although the latter alone does not define functional impairment in children of Sprengel's shoulder, the main physical disability, such as the inability to wear clothes, arises due to the restriction in shoulder abduction. According to the Cavendish system of grading, which has been universally accepted, Sprengel's shoulder can be divided into four grades, depending on the level of the shoulders and visibility of the hump (Fig. 1). The operative indications in our patients were severe restriction of range of abduction at the shoulder, interfering with activities of daily living or a cosmetically displeasing deformity, or both.

Under general anaesthesia, a midline longitudinal incision was made from the midcervical region to the lower thoracic vertebra. The trapezius and rhomboid muscles were detached from their origins on spinous processes. Any omovertebral bone or fibrous band extending from the

superomedial angle to the cervical spine was excised (Fig. 2). After ensuring mobility of the scapula, the bone was displaced caudally to align the scapular spine to the level of the contralateral scapula. The scapula was anchored with heavy absorbable sutures at the tenth thoracic vertebra. The trapezius and both rhomboids were resutured at a lower level. Postoperatively, the limb was immobilised in a Velpeau sling and range of motion exercises begun at 3 weeks postoperatively.

Outcome assessment was done by measuring the range of abduction at the shoulder joint at final follow up and compared to a similar measurement made preoperatively. Similarly, Cavendish grades at final review were compared to those before surgery. The lowering of scapulae achieved was estimated by the vertebra corresponding to the level of the spine of the scapula in an anteroposterior radiograph showing both shoulders. Any associated malformation was sought and its impact on the outcome was assessed.

Fig. 2 Omovertebral bar being excised as part of the surgical procedure. The bar is marked by arrows

The effect of age at the time of surgery and preoperative severity on the final cosmetic and functional outcome was also evaluated.

Statistical analysis was done using SPSS v.19 software (SPSS Inc., Chicago, IL, USA). The Wilcoxon signed-rank test was used for the paired analysis of preoperative and final values of Cavendish grades and range of abduction. Impact of omovertebral bar and associated malformations on the end result was evaluated by the Mann–Whitney test. Pearson's correlation coefficient was used to evaluate the relationship between age, cosmetic enhancement, functional improvement and scapular lowering achieved. A p -value <0.05 was considered to be significant.

Results

Seven girls and five boys were operated at a mean age of 5.58 years (range 1–11 years). The number of left and right shoulders operated was six of each. The mean follow up was 31.8 months. Four patients had an associated anomaly—two had scoliosis with hemivertebra, one showed rib anomalies and one had bilateral polydactyly of the feet.

The mean arc of abduction in the shoulder of the involved side was 115.8° (70 – 160°), which increased to 153.3° (120 – 170°) at final follow up, with an improvement of 37.5° . The increase in the range of motion was significant on statistical analysis ($p < 0.0005$, Wilcoxon signed-rank test), depicting a good functional outcome with the procedure. The mean preoperative Cavendish grade for cosmetic evaluation was 3.17 (range 2–4), which decreased to 1.25 (range 1–2) at final review (statistically significant, $p < 0.0005$, Wilcoxon signed-rank test). Cavendish grade improvement and increase in abduction had a strong

Fig. 3 Scatter diagram showing the relationship between the improvement in shoulder abduction ($^\circ$) and cosmetic improvement (Cavendish grade)

Fig. 4 Preoperative clinical radiograph showing a Cavendish grade 4 deformity

positive association ($r = 0.713$, Spearman correlation coefficient, Fig. 3). A typical clinically good result is shown in Figs. 4 and 5.

The Woodward procedure achieved a mean lowering of 2.1 vertebral levels at the latest review as compared to the preoperative radiographs in our series (Figs. 6 and 7). However, there was no correlation between an increase in abduction and lowering of the scapula achieved ($r = 0.131$, Spearman correlation coefficient).

The effect of age on the final outcome was evaluated using correlation studies. It was found that age had a negative effect on the functional outcome ($r = -0.55$) and cosmetic outcome ($r = -0.11$). The effect of age was more pronounced on functional improvement, implying that surgery at older age does not assure an increased abduction at the shoulder but a good cosmetic outcome can be expected.

33.3 % (4/12) patients had an associated omovertebral bar which was excised. Clavicle osteotomy was done in one patient (indication was comparatively higher age, being 11 years) to ensure adequate mobilisation of the scapula and prevent any neurological complication. Comparison of outcome was done in two subsets of patients with or without an omovertebral bar. It was found that the presence or absence of the bar had no impact on the eventual outcome.

Fig. 5 Follow up clinical photograph at 4.5 years follow up with the Cavendish grade 4 deformity being reduced to grade 1

Fig. 6 Preoperative anteroposterior radiograph showing elevated scapula on the left with spine of the scapula at the level of the sixth cervical vertebra. Also seen is the omovertebral bar (marked by arrows)

The patients and parents were satisfied with the outcome of the surgery. None of the patients developed infection or brachial plexus injury in our series.

Discussion

Congenital elevation of the scapula was first described by Eulenberg [11] in 1863, but the eponym Sprengel's shoulder was coined by Kollicker after a description of four cases in 1891 [12]. Sprengel's shoulder arises as a result of arrest of normal caudad migration of the scapula during the

Fig. 7 Follow up radiograph at 4.5 years follow up of the same patient with spine of the scapula at the level of the first thoracic vertebra, thus, achieving a lowering of two levels. Note the derotation of the scapula

organogenesis phase of intrauterine life. In addition to the difference in height, Horwitz [1] identified four major differences that an elevated scapula has from its counterpart; changed relation of diameters, curving of the superior border or bending forward of the supraspinous portion, prolongation or rounding of the superior median angle, and presence of exostosis or articulation with the vertebral column. The incidence of the latter is highly variable, ranging from 25 % to as high as 88.9 % [1, 5]. We encountered such a bar in 33.3 % (4/12) of our patients. It is recognised that tethering by the omovertebral bar is an additive deforming force during scapular development [13], besides many other factors. Removal of this connection is a routine part of various kinds of surgeries done for elevated scapulae. The bar should be dissected in its full dimensions, and the excision may entail even exposure of the dura mater; however, we did not encounter exposure of the dura mater in any of our patients. We found, upon statistical comparison, that its presence does not alter the results obtained with regard to cosmesis and shoulder abduction after the Woodward procedure.

The cosmetically displeasing deformity is usually accompanied by a decrease in abduction of the ipsilateral shoulder. Severity grading done by the Cavendish system is a widely used classification method. In our series, 66.6 % (8/12) were grade 3, 25 % (3/12) were grade 4 and 8.4 % (1/12) were grade 2.

Various surgical procedures have been described for elevated scapulae, all offering a reasonable and comparative outcome. However, the preferred method [14] has been the Woodward procedure, since it does not require a postoperative spica cast for suture anchor and entails a

Table 1 Demographics and results of patients of Sprengel's deformity treated by the Woodward procedure by different authors

Study author(s)	Average age (years)	Number of cases	Increase in function	Improvement in cosmesis (Cavendish grade)
Woodward [8]	8.1	9	32°	–
Crha and Gál [15]	7	18	46°	0.67
Grogan et al. [16]	6.5	20	37°	–
Wu et al. [17]	4.7	9	78°	2.23
Carson et al. [4]	9	11	29°	–
Ross and Cruess [18]	–	17	35°	–
Present study	5.6	12	37.5°	1.92

much lower risk of bleeding and brachial plexus injury. The mean patient age at the time of undergoing surgery in our study was 5.58 years, which is on the lower side than the age at which cases have been operated by the Woodward procedure in various series of patients with Sprengel's shoulder [4, 8, 15–18]. The improvement in shoulder abduction and the Cavendish grade at final follow up in our study was quite comparable to other series managed by the Woodward procedure (Table 1). We found that functional and cosmetic outcome had a positive correlation, an inference not previously reported. We found that, as the duration of follow up increases, further cosmetic and, especially, functional improvement is seen. However, since the absolute values were not measured over the range of review due to the retrospective nature of the study, a concrete statement cannot be made to this effect.

We achieved an average scapular lowering of 2.1 vertebral bodies. Satisfactory lowering has been reported by various authors using the Woodward procedure. However, a significant lowering achieved may not assure a good increase in the range of motion.

Opinion is divided regarding the optimal age for correction. Surgical correction before 3 years of age is technically more demanding than in an older child [3], but as age increases, the results tend to diminish. We found that, as age increases, the functional outcome deteriorates but cosmesis does not. The same was found by Borges et al. [19], who noted, in two older children, that a modified Woodward procedure improved appearance but not function.

Brachial plexus palsy remains one of the most dreaded complications of the procedure. Many series have

documented postoperative neurological involvement, either transient or permanent, after the Woodward procedure. However, fortunately, we did not encounter any such occurrence in our study. We feel that overzealous attempts at correction should be particularly avoided in order to prevent the same. The role of clavicular osteotomy in the prevention of brachial plexus palsy remains debatable [16, 20]. We performed the same in only one child who was 11 years old at the time of surgery. Although there is no direct evidence to suggest that clavicular osteotomy prevents neurological complications, it was done only as a prophylactic measure. It also helps to mobilise the scapula more in older children, where the joints are not supple enough to allow a good surgical descent. Other complications which may plague the procedure are winging of the scapula, infection and unsightly keloid-ridden scar marks.

A small sample size is a potential limitation of our study. However, considering the rarity of the disorder, 12 cases present an adequate sample size to be able to draw meaningful inferences.

Conflict of interest The authors declare they have no conflict of interest.

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