Sprengel Deformity: Morphometric Assessment and Surgical Treatment by the Modified Green Procedure

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Background: We evaluated the pathologic anatomy of the Sprengel deformity using radiographs, 3-dimensional computed tomography (3D-CT), and magnetic resonance imaging (MRI), and reviewed our results of the modified Green procedure. 

Methods: Between 2002 and 2009, 23 scapulae in 22 patients were treated. The average age at the time of surgery was 3.4 years (range, 1.9 to 7.1 y). The outcome of surgery was assessed on the basis of changes in shoulder abduction and the radiographic findings using Cavendish classification, Rigault classification, scapular elevation, and medialization. Preoperative appearance was classified as grade III in 13 cases and grade IV in 10 cases according to the Cavendish classification, and as grade II in 4 cases and grade III in 19 cases according to the Rigault classification. Using 3D-CT, we assessed the height to width ratio on the scapular posterior view, the superior displacement ratio and rotational difference on the trunk posterior view, and the anterior curvature of the supraspinous portion on the scapular medial view. 

Results: The average follow-up postoperative period was 4.4 years (range, 2.2 to 8.7 y). Postoperatively, the shoulder abduction improved well, with a mean improvement of 63 degrees. Improvement of at least 1 Cavendish grade and 1 Rigault grade became associated with his name. This congenital deformity arises from interruption in the normal caudal migration of the scapula during the ninth to the 12th week of the embryologic period, resulting in an elevated and hypoplastic scapula. Scapular adduction and medial rotation, prominence of the upper corner of the scapula, abnormalities in the cervicothoracic vertebrae or thoracic rib cage, and changes in the clavicle position have also been detected. In the Sprengel deformity, there is restriction of the scapulothoracic motion, which restricts shoulder abduction. Surgical treatment for patients with the Sprengel deformity is indicated when patient and family expectations are either improved cosmesis or improved function. 

Conclusions: 3D-CT and MRI were helpful in evaluating the pathologic anatomy of the deformity and in planning the surgery. The modified Green procedure provided successful functional and cosmetic results. 

Level of Evidence: Level IV. 

Key Words: Sprengel deformity, congenital elevation of the scapula, modified green procedure, scapuloplasty, Klippel-Feil syndrome, omovertebral bone

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Eulenburg is credited as the first person to describe congenital elevation of the scapula, in 1863. Sprengel described 4 cases of this condition, and the condition became associated with his name. This congenital deformity arises from interruption in the normal caudal migration of the scapula during the ninth to the 12th week of the embryologic period, resulting in an elevated and hypoplastic scapula. Scapular adduction and medial rotation, prominence of the upper corner of the scapula, abnormalities in the cervicothoracic vertebrae or thoracic rib cage, and changes in the clavicle position have also been detected. In the Sprengel deformity, there is restriction of the scapulothoracic motion, which restricts shoulder abduction. Surgical treatment for patients with the Sprengel deformity is indicated when patient and family expectations are either improved cosmesis or improved function. 

The purpose of the present study was to review our clinical and radiographic results after the use of the modified Green procedure for the Sprengel deformity. Three-dimensional computed tomography (3D-CT) and magnetic resonance imaging (MRI) were used preoperatively to evaluate the pathologic anatomy of the deformity, including malposition and dysplasia of the scapula and presence of omovertebral connections, and to plan surgical treatment. 

METHODS 

Between 2002 and 2009, 23 scapulae in 22 patients were treated with the modified Green procedure at our institution. The details of the 22 patients are shown in Table 1. The patients included 13 boys and 9 girls. The affected scapulae were bilateral in 1 case and unilateral in 21 cases, with the left scapula affected in 16 and the right in 5. Surgery was indicated if the deformity and shoulder...
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3D-CT indicates 3-dimensional computed tomography; BPP, brachial plexus palsy; CO, clavicular osteotomy; HS, hypertrophic scarring; KFS, Klippel-Feil syndrome; LOC, loss of correction; RRD, radial ray deficiency; SB, spina bifida; SC, scoliosis; SW, scapula winging.
impairment were severe. The average age of the patients at the time of surgery was 3.4 years (range, 1.9 to 7.1 y).

We performed the modification of the Green procedure under general anesthesia with deliberate hypotension in this study. A curved incision is made from the middle of the spine of the scapula to the superomedial corner and then descending along the scapular vertebral border distally to the inferior angle of the scapula. The original Green procedure consists of extraperiosteal detachment of the medial and superior scapular muscles, extraperiosteal resection of the omovertebral connection, excision of the supraspinous portion of the scapula, lowering positioning of the scapula, and reattachment of the muscles to the lowered scapula. The position is then secured by a percutaneous wire or spring system and is maintained by a plaster jacket. In the modified Green method used in this study, the scapula was preserved in its lowered and furthermore laterally rotated position by rejoining the detached muscles, in particular the serratus anterior, with care to prevent postoperative scapular winging.9 The inferior angle of the scapula was placed into the pocket created beneath the latissimus dorsi muscle, eliminating the need for a traction system.9 Clavicular osteotomy was performed in the supine position before the modified Green procedure in 2 cases to reduce the risk of brachial plexus palsy. The clavicle was released by removing its middle portion, morcellating the removed segment, and replacing it in its periosteal sheath.10,11 To prevent unsightly scarring, we used an intradermal buried suture technique with polydioxanone monofilament (PDS2; Ethicon, Somerville, NJ) for closure of the wound. Additional skin suturing was not used, to avoid scarring of suture holes along the incision. The patients applied paper tape to their scars for 3 to 4 months after suture removal to eliminate scar tension and to prevent hypertrophic scarring. Postoperatively, we held the arm in a Velpeau bandage for 2 to 4 weeks, and the patient began active exercise to strengthen the abductor muscles.

The outcome of surgery was assessed on the basis of changes in shoulder abduction and the radiographic findings evaluated using Cavendish classification,12 Rigault classification,13 scapular elevation,4,14 and medialization.4 Cavendish12 classified the deformity into the following 4 grades: grade I (very mild), the shoulder joints are level, and the deformity is invisible when the patient is dressed; grade II (mild), the shoulder joints are level, but the deformity is obvious when the patient is dressed; grade III (mild), the shoulder joints are level, but the deformity is obvious when the patient is dressed; as a lump in the web of the neck; grade III (moderate), the shoulder joint is elevated 2 to 5 cm with obvious deformity; grade IV (severe), the affected shoulder is elevated, meaning that the superior angle of the scapula is near the occupit. Fourteen cases were classified as Cavendish grade III and 9 as grade IV preoperatively. Rigault et al13 classified the deformity into the following 3 grades: grade I, the superomedial angle is below the T2 but above the T4 transverse process; grade II, the superomedial angle is located between the C5 and the T2 transverse process; grade III, the superomedial angle is above the C5 transverse process. Four cases were classified as Rigault grade II and 19 as grade III preoperatively. Scapular elevation was measured by the Spearman nonparametric correlation test. We tested the correlation between the superior displacement ratio and the rotational difference by the Spearman nonparametric correlation test.

RESULTS

Spina bifida was found in 21 cases, Klippel-Feil syndrome in 19 cases, scoliosis in 4 cases, and radial ray deficiency in 2 cases. Omovertebral bone connection was found in 20 cases. One had bony, 13 had cartilaginous, and 6 had fibrous connections with associated scapulae. In 1 patient with bilateral deformities (Fig. 3), omovertebral bones were not ossified and were not seen on radiographs or CT scans. MRI identified bilateral omovertebral cartilages that connected to the scapulae (Fig. 3B).

On preoperative 3D-CT, the mean height to width ratio of the affected scapulae was 1.22 (range, 1.04 to 1.41), and that of the contralateral scapulae was 1.52 (range, 1.30 to 1.65). The mean height to width ratio of the affected scapulae was significantly smaller than that of the contralateral scapulae (P < 0.001). The mean superior displacement ratio was 0.23 (range, 0.02 to 0.52), and the rotational difference was 27 degrees (range, 9 to 43 degrees). There was an inverse relationship between the superior displacement ratio and the rotational difference (P = 0.011). Anterior curvature of the supraspinous portion was seen in all patients.

The mean operative time and blood loss at surgery were 125 minutes (range, 77 to 175 min) and 16 mL (range, 5 to 35 mL), respectively. No blood transfusion was received by any patient. The average follow-up postoperative period was 4.4 years (range, 2.2 to 8.7 y). One patient had a loss of correction of the deformity at the time of first outpatient visit after surgery. The modified Green procedure was performed again for this
patient. Complete brachial plexus palsy occurred postoperatively in 2 cases associated with Klippel-Feil syndrome. Both patients had fully recovered within 4 months. No clavicular osteotomy was performed in the 2 cases. Surgical scarring was hypertrophic in 8 patients. Unsightly spreading of the scar was seen in the proximal half of the incision. In 1 case, the parents complained about the cosmetic appearance of the scar formation, and scar revision was performed in this patient. Postoperative scapular winging was observed in 3 cases.

The differences in preoperative and final follow-up shoulder abduction, Cavendish grade, Rigault grade, scapular elevation, and medialization were statistically significant ($P < 0.001$). Preoperatively, the mean shoulder abduction was 88 degrees (range, 70 to 100 degrees). At final follow-up, the mean shoulder abduction was 151 degrees (range, 130 to 170 degrees). The shoulder abduction improved in all cases, with a mean improvement of 63 degrees. Improvement of at least 1 Cavendish and Rigault grade was attained in all cases. At the final follow-up, the 13 preoperative Cavendish grade III cases were classified as grade I in 8 cases and II in 5 cases, and the 10 preoperative grade IV cases were classified as grade II in 5 cases and III in 5 cases. The 4 preoperative Rigault grade II cases were classified as grade I in 4 cases, and the 19 preoperative grade III cases were classified as grade I in 18 cases and II in 1 case. Preoperatively, the mean scapular elevation was 32 mm (range, 21 to 45 mm) and medialization was 23 mm (range, 15 to 34 mm). At final follow-up, the mean scapular elevation was 15 mm (range, 0 to 32 mm) and medialization was 31 mm (range, 19 to 39 mm).

DISCUSSION

In patients with the Sprengel deformity, the affected scapula has been described as hypoplastic with a decreased height to width ratio and occasionally with an anteriorly curved supraspinous portion.14-17 The mean ratio of affected scapulae in our study was 1.22 (range, 1.04 to 1.41) and was significantly smaller than that of contralateral scapulae at 1.52 (range, 1.30 to 1.65). Ogden and Phillips17 reported the mean height to width ratio in 35 normal cadaver scapulae in the age range from 0 to 14 years was 1.49 (range, 1.42 to 1.56). The decreased height to width ratio of the affected scapulae was due to the decreased height at the anteriorly curved supraspinous portion and the increased width at the convex medial border in the infraspinous portion, which appeared to be related to the omovertebral tethering. Anterior curvature of the supraspinous portion was seen in all cases, and omovertebral bone was found in 20 of 22 cases in our series. The height to width ratio of the contralateral scapulae in our series was not smaller than that of the normal scapulae in the study of Ogden and Phillips.17
Meanwhile, the height to width ratio of the contralateral scapulae was slightly smaller (mean 1.40; range, 1.32 to 1.58) in the study conducted by Cho et al., who suggested that the morphogenesis of the contralateral scapula, even if fully migrated, may be affected and dysplastic. There was a significant inverse correlation between the superior displacement ratio and the rotational difference, which was compatible with findings reported by Cho et al. Thus, in cases with little medial rotational deformity, the glenohumeral joints remain in a marked elevated position compared with the contralateral scapula (Fig. 4). Meanwhile, in cases with marked medial rotational deformity, the glenohumeral joints remain in a slightly elevated position compared with the contralateral scapula (Fig. 5). Accordingly, the inferior medial angle of the scapula was recommended to be used as the radiographic reference point for preoperative and postoperative measurements, and it was also used in the present study.

In our series, omovertebral bone connection was found in 20 of 23 cases (87%). Cho et al. described the role of the omovertebral bone connection in the pathomechanism of the Sprengel deformity. They hypothesized that a “pulling force” during embryonic development acts on the normal scapula in caudal and lateral directions, but the presence of omovertebral bone associated with the Sprengel deformity causes the malpositioning of the scapula. Omovertebral connections were tethered at the convex medial border of the infraspinous portion of the scapula, and the tethering medial to the migrating

**FIGURE 3.** Bilateral Sprengel deformity (case 22). A, Preoperative radiograph at 3.4 years showing marked elevated and medially rotated deformity. B, Coronal T2-weighted multiple-echo data image combination (MEDIC) sequence magnetic resonance imaging (TE/TR, 21/581 ms) showing bilateral unossificated omovertebral cartilages (arrowheads) connected to the bilateral scapulae. The modified Green procedure was performed at 3.4 years on the right scapula and at 3.8 years on the left scapula. C, At 7.1 years, preoperative elevation and medial rotation both remained improved.

**FIGURE 4.** Trunk posterior 3-dimensional computed tomography view of left Sprengel deformity (case 2). The affected left inferior angle (I), superomedial angle (S), and glenohumeral joint (G) are elevated with little rotation, although omovertebral bone (arrowheads) has a cartilaginous connection to the infraspinous portion of the scapula.
force affected the rotational torque, resulting in marked rotation and an inferiorly faced glenoid cavity. However, there was an exceptional case in our series, in which the scapula remained in the cephalic position with little rotation, although omovertebral bone connected to the infraspinous portion of the scapula (Fig. 4). The omovertebral tethering did not affect the rotational torque in this case.

Many authors refer to Cavendish classification for surgical indication. Although no improvement or worsening has been reported in untreated grade I and II cases, surgery is recommended in grade III and IV deformities.4,6 However, Cavendish classification remains subjective and inaccurate, as it is based on morphologic and aesthetic criteria.18 Rigault classification is objective for surgical indication18 and was also used in this study.

Various procedures have been described, the goal of which is to improve the cosmetic appearance and shoulder function, including scapular lowering by detaching muscles at their insertion points.4,8,9,18,19 scapular lowering by detaching muscles at the spinal origin,10,14,16,20,21 scapular osteotomy,22,23 partial resection technique,24,25 and partial scapulectomy.26 The Green8 and Woodward21 procedures are commonly performed techniques. The Woodward procedure is more widely used than the Green procedure,7 but the surgeon’s choice between the 2 techniques is often based on personal preference, and the final results are comparable.7,19 We used the modified Green procedure because it provides a good visualization of the superomedial scapular angle, which is generally anteriorly curved and lying on the thorax.18 The modified Green procedure also provides better access to the acromiothoracic vascular nervous pedicle, which must be carefully protected before bony resection.18 Another advantage of this method is that the lever arm relative to the scapular center of rotation is enlarged by intraoperative exposition of the scapula and by muscular reinsertion at a different site on the scapula.18 By use of this method, the elevated and medially rotated scapula is able to be both lowered and laterally rotated.4,18 Successful functional and cosmetic results were obtained in this study. At the final follow-up, the Cavendish and Rigault grades were improved in all cases.

We recommend that the modified Green procedure be performed as soon as possible after 2 years of age, because soft tissues will be more flexible and better correction can be obtained, and early operation allows for continued growth and development. However, some authors3,4,14 reported that in patients younger than 3 years, identification of the anatomy is more difficult and the operation becomes technically more demanding. We performed the modified Green procedure in 12 patients younger than 3 years. Deliberate hypotensive anesthesia was applied in all cases, which reduced blood loss and provided good visualization during surgery. The mean blood loss at surgery was 16 mL, and no blood transfusion was needed by any patient.

We agree with other authors9,12 that patients who are more than 6 years old are not suitable for the scapular displacement procedure. All of our patients except 1 were operated on when they were under the age of 6. The patient operated on at the age of 7.1 years had transient brachial plexus palsy after surgery. In older patients, sufficient correction becomes difficult, and brachial plexus palsy may occur because their soft tissues are less flexible compared with that at a younger age.4

We had 2 patients with brachial plexus palsy after surgery, both of whom had completely recovered within 4 months. Clavicular osteotomy was not combined with the modified Green procedure in these 2 cases. Previous authors18,20,25 have also reported transient brachial plexus palsy after surgery. Clavicular osteotomy has been advocated to prevent brachial plexus palsy.4,7,10,18 The issue of whether clavicular osteotomy should be performed remains controversial. Some authors4,7,10 performed clavicular osteotomy in all cases, whereas others9,19,20 did not perform clavicular osteotomy in any of the cases. After the experience of transient brachial plexus palsy, we decided to perform clavicular osteotomy preoperatively in patients who are more than 4 years old and/or who are associated with severe Klippel-Feil syndrome. Somatosensory-evoked potential monitoring is widely used for monitoring spinal cord function during spine surgery, and, recently, it has been used to monitor brachial plexus function.27 Shea et al27 used somatosensory-evoked potential to monitor brachial plexus function during a Woodward procedure. They identified brachial plexopathy during the procedure and were able to appropriately modify the position of the scapula. Additional neuromonitoring, including electromyography for neurotonic discharges, may also be considered.27

Postoperative management varies. Some authors14,28 reported that the shoulder is placed in a shoulder immobilizer for 6 weeks. Others9,18 reported that the shoulder is immobilized for 4 weeks. Bellemans and Lamoureux19 described that gentle active and passive exercises are started 2 days after surgery, and the arm is...
held in a sling for 3 weeks. Ahmad recommends immediate postoperative mobilization with physiotherapy. In 1 patient in this study, the deformity was recurrent at the time of the first outpatient visit after surgery. We held the arm in a Velpeau bandage for 2 weeks but allowed active exercise for the patient. In the next 6 cases after the recurrence of the deformity, we immobilized the shoulder in a Velpeau bandage for 3 to 4 weeks, after which the patient began active exercise to strengthen the abductor muscles.

In terms of keloid formation risk and surgical scarring, better results were reported using the Woodward procedure compared with the Green procedure. A curved incision is made from the middle of the spine of the scapula to the superomedial corner and then descending along the scapular vertebral border distally to the inferior angle of the scapula in a modified Green procedure. Meanwhile, a midline incision is made from the spinous process of the fourth cervical spine to the ninth thoracic spine in the Woodward procedure. The incision in the modified Green procedure is shorter, but it is more conspicuous than that in the Woodward procedure. However, unsightly scar formation was reported in 64% after the Woodward procedure in a study by Carson et al, and in only 22% after the modified Green procedure in a study by Gonen et al. Unsightly scar formation was observed in 8 cases (35%) in our series, which was comparable to previously reported prevalence (22% to 64%). As described in the previous reports, the unsightly spreading of the scar occurred in the proximal half of the incision.

Postoperative scapula winging was seen in 3 cases. Borges et al described that decreasing the width of the scapula by excision of the convex medial border prevents postoperative scapula winging. We did not excise the convex medial border in the infraspinous portion in our series, which may have caused postoperative scapula winging. Gonen et al reported that scapula winging is the result of muscle imbalance or of failure to reattach the serratus anterior to the scapula. Bellemans and Lamoureux presented the modified Green procedure that includes preserving the serratus anterior, but the scapula is repositioned merely by rotation rather than by lowered translation.

One of the limitations of this study is that the number of cases is limited compared with some larger series. Another limitation is that the follow-up period after the modified Green procedure is not long enough to reveal all of the possible outcomes of using the procedure. Cavendish mentioned that improvement in function is uncertain and that cosmetic appearance often deteriorates during a period of rapid growth of the neck. However, successful long-term functional and cosmetic results were provided after the modified Green procedure in the studies conducted by Gonen et al and Bellemans and Lamoureux. We believe that the modified Green procedure is a safe and effective technique in patients with severe Sprengel deformity at the age of 2 to 6 years, although this procedure may have to be combined with clavicular osteotomy in older patients and/or when the deformity is associated with severe Klippel-Feil syndrome.

REFERENCES

