

Overview of Sprengel Deformity in Children

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Abstract

Sprengel deformity is a congenital condition characterized by abnormal development and elevation of the scapula (shoulder blade). The severity can vary considerably from being nearly invisible when covered by clothing, to being raised more than 5 cm, with webbing of the neck. Signs and symptoms may include a lump behind the base of the neck and limited movement of the shoulder or arm. The condition may also be associated with other skeletal (bone or cartilage) or muscular abnormalities. Sprengel deformity usually occurs sporadically for no apparent reason but autosomal dominant inheritance has been reported. The condition is caused by a disruption in the normal development and movement of the scapula during early fetal development (probably between the 9th and 12th weeks of gestation). Classification may be according to the Cavendish clinical classification or the Rigault classification on radiographs. Treatment usually involves physical therapy, but severe cases may require surgery. The ideal time for surgery is between 3 and 8 years of age. Various surgical treatments are described in the literature to improve the cosmetic appearance and function of the scapula; these include: Woodward, Putti, Putti, Delchef, Schrock, McFarland, Green, Allan, Woodward. Schrock, Green, Mears and Woodwards have described various modifications to lower the scapula. Following surgery, the patient's shoulder is immobilized to promote healing and reduce any pain that may occur. Reduction of deformity and allowing normal shoulder motion are the goals of physical therapy.

Keywords: Sprengel's Deformity; Woodward Procedure; Abduction of the Shoulder; Omovertebrae; Klippel-Feil Syndrome

Introduction

Michel Mortiz Eulenberg described this malformation as high-grade dislocation of the scapula (hochgradige dislocation der scapula) in 1863 in three patients [1]. About 22 years later in 1863, Willet and Walsham were the first to describe the omovertebral bone and gave a comprehensive anatomical description, method of resection, and reported good results (Figure 1). Sprengel's malformation is associated with many other congenital malformations. Sprengel himself in 1891 illustrated this malformation in four cases and hence its name. Howitz in 1908, Fairank in 1914, and Grieg in 1924 reported a few cases.

The exact cause of this congenital anomaly or the presence of omovertebral bones is unknown, but it is clear that during the embryonic period, disruption of scapular migration occurs from the level of the embryonic limb buds opposite C5, which also causes failure of normal bone and soft tissue development in the shoulder girdle [2-4]. It is hypothesized that all these tissues and the scapula are of mesodermal



Figure 1: Anapathology of Sprengel deformity.

origin and develop during the same embryonic period. Therefore, the failure may result in a number of associated anomalies [5]. Depending on the severity, the anomaly may be apparent at birth or manifest later in childhood. Sprengel's anomaly is a congenital condition in which the scapulae are elevated due to failure to descend during embryonic development. It is a rare anomaly characterized by one scapula being higher than the other. This complex anomaly is associated with scapular agenesis and hypoplasia of the muscles surrounding the scapula. Although rare, this is the most common deformity of the shoulder girdle.

Although both shoulders can be affected, the left shoulder appears to be affected more often. Among the sexes, the condition is more common in women with a female to male ratio of approximately 3:1. The condition is often associated with conditions such as Klippel Feil malformation (about one-third have Sprengel malformation), scoliosis, omovertebral bar, spina bifida, torticollis, and clavicle abnormalities.

The dysplastic scapula is located higher than normal in the neck or upper chest. It is smaller than normal in the vertical plane and appears larger horizontally. The inferior angle is rotated medially and proximally, causing the glenoid to point downward.

Limitation of abduction and shoulder aesthetics are the main concerns. In this condition, several procedures have been described to correct the deformity. Among the procedures described, the modified Woodward procedure has been found to be particularly beneficial. However, few studies have described the functional outcomes of surgical correction of Sprengel deformity. In this study, we evaluated the outcomes of a modified Woodward procedure that included separation of the trapezius roots, excision of the trapezius and levator scapulae from the spinous process, posterior displacement of the levator scapula after resection of any omovertebral or fibrous connections on the scapula, and excision of the superior medial angle of the scapula. Hung NN. Pediatric Orthopedics in Guidance The separated scapulae and muscles were repositioned at a lower level with heavy sutures and were gradually rehabilitated postoperatively using slings and exercises. Cosmetic evaluation was performed using the Cavendish score and radiographic parameters according to the Rigault score before and after surgery to assess the severity of the deformity, the position of the scapula relative to the cervical spine before and after surgery. Radiographic evaluation also included measuring the ratio of upper displacement and the height-to-width ratio of the scapula in the posterior scapular view from preoperative images such as chest radiographs and comparing with postoperative images.

Functional and subjective evaluation were also performed using the PODCI (Pediatric Outcomes Data Collection Tool) and the Simple Shoulder Test (SST).

Embryology scapula

Congenital prominence of the scapula, also known as Sprengel's malformation, is the most common congenital anomaly of the shoulder girdle [6,7]. The pathophysiology of Sprengel's malformation is particularly related to the embryology of the upper limb. Scapula

formation occurs during the embryonic period through germinal differentiation between the third and eighth weeks of gestation. The scapula is located at the level of the fourth and fifth cervical vertebrae at this time [7]. Continuous cell signaling from surrounding tissues is required for mesoderm differentiation into the axial skeleton and limb bones. Programmed mesenchymal cell pathways are required for scapula development. Multiple cell signaling molecules such as bone morphogenetic proteins and fibroblast growth factors induce multipotent mesenchymal cells to differentiate into skeletal tissues [8]. A specialized group of mesenchymal cells, the apical ectodermal crest, present at the periphery of the limb bud, directs lower limb development. The development and growth of the scapula is guided by multifaceted cell signaling pathways, which in turn direct the development and growth of surrounding muscles, bones, and nerves. The scapula develops together with the upper limb in the upper back/lower neck with the humeral bud during the 5th week of gestation and descends to the 2nd - 8th thoracic vertebrae, reaching its final anatomical position by the 12th week of gestation (final position at the level of the scapular spine at the 12th week. The pathology in Sprengel's malformation may be a continuation of the fetal scapular shape. It has been described that the deformed scapula is smaller in longitudinal diameter than transverse diameter, with the supraspinous portion curved forward with an elongated superior angle between the scapula [11,12]. The anterior curvature of the supraspinatus with an elongated superior medial angle of the scapula is due to a smaller deformed scapula whose transverse diameter exceeds its longitudinal diameter.

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The scapula normally descends to its thoracic position around the twelfth week of gestation, forming from the paraxial mesoderm at approximately the level of the fourth or fifth cervical vertebrae.

The normal post-migration position of the scapula is between the levels of the second and eighth posterior ribs. Surrounding structures are also affected, requiring a normal scapula for development. Sprengel deformity occurs due to problems with limb bud formation. The most obvious manifestation is a prominent scapula, seen in conjunction with hypoplasia of the scapula and surrounding musculature. The scapula bone formation is via intramembranous ossification. The pectoralis major, trapezius, rhomboids, levator scapulae, serratus anterior, and latissimus dorsi are the muscles that act on the scapula, and varying degrees of involvement of these muscles are seen in congenital scapular prominence [13]. The trapezius has been reported to be atrophied in cases of acquired scapular prominence [14]. The trapezius attaches along the medial border of the scapula and resists the upward forces of the levator scapulae and rhomboids.

Some of the proposed hypotheses for the cause of these syndromes include the inheritance of an autosomal dominant, vascular lesion arising from the subclavian artery [13,15,16]. Most cases are sporadic and the cause is unknown. It is unclear whether the inheritance of autosomal or vascular lesions is the cause of Sprengel anomaly, but they may not be mutually exclusive. Other anatomic findings include the omo-vertebral bar, which articulates with the spine; and these may extend from the supra-medial angle of the scapula or the upper third of the medial border of the scapula up to the transverse process of the cervical vertebrae (one of the fourth to seventh vertebrae). In addition, various types of disruptions in limb bud formation and differentiation can be noted, including hypoplastic thumbs, hypoplastic clavicles and hemimelias, rib-vertebral defects (spina bifida and kyphoscoliosis), and underdevelopment of the thoracic girdle bones (clavicles, humerus) and muscles (pectoralis major, trapezius) that may coexist. Other embryonic abnormalities include Poland anomaly, Klippel-Feil syndrome, and Möbius syndrome [8]. Due to the cooperative development of the skeletal system, such conditions result in a variety of phenotypic abnormalities.

Sprengel deformity

Sprengel deformity is the most common congenital anomaly in children [17] in which the scapula is positioned too high, mainly on one side. Usually during fetal development, the scapula moves down the back to its normal position. These children will have cosmetic problems, limited mobility, especially in abduction and forward flexion of the affected shoulder. Associated anomalies such as Klippel-Feil syndrome and scoliosis often coexist, and affected children are at increased risk of renal disease.

It is defined as a rare congenital anomaly. During embryonic development, it causes the scapula to fail to descend normally, from its position in the neck to its normal position in the posterior chest. The affected scapula is smaller and more cephalad than the normal scapula, characterized by internal rotation of the lower portion of the affected scapula and elevation [11,18].

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Shoulder deformity and limited mobility are due to regional muscle atrophy or atrophy. The deformity is usually unilateral, but can also be bilateral [10,18-20]. Sprengel deformity can be isolated or part of a syndrome. In about one-third of patients, the affected scapula is attached to the cervical spine by omovertebral bone (cartilage or fibrous tissue), which, when present, makes shoulder extension beyond 90 degrees virtually impossible [12,21,22]. Sprengel deformity is characterized by a prominent scapula, asymmetrical shoulder contour, and limited shoulder range of motion. It is caused by a variable arrest of the scapula during intrauterine development.

Sprengel deformity may also occasionally occur as part of Klippel-Feil syndrome (in 30% of cases) or may be associated with other spinal and cranial anomalies or rib absence [5,11,18,23]. The upper part of the scapula may protrude and curve forward over the top of the chest, resulting in a clavicle deformity. The shoulder muscles are hypoplastic, sclerotic, or even absent in most cases, and weakness of the serratus anterior muscle may cause a bowed scapula [21,24]. In 20 - 30% of patients, an omovertebral "bone", consisting of bone, cartilage, or fibrous tissue, is found between the upper corner of the scapula and the spinous process of the cervical vertebrae.

Epidemiology

Sprengel deformity is the most common congenital anomaly of the shoulder girdle. In general, the condition has a female predominance of 3:1 [2,23] and usually affects one shoulder; however, bilateral involvement has been described [12,25].

This usually painless condition causes partial impairment of shoulder mobility, especially scapulothoracic abduction and rotation, resulting in functional disability and disruption of normal cosmetic appearance. It is not known whether this altered anatomy leads to early degenerative changes in the shoulder joint. The primary goal of surgical treatment of Sprengel deformity is to improve shoulder function as well as cosmetic appearance.

Etiology

The condition is sporadic. Rarely, it exhibits an autosomal dominant inheritance pattern, known as Corno disease [26,27].

Clinical feature

Children often present with complaints that the affected scapula is higher than normal, approximately 2 to 10 cm. The scapula appears larger than normal horizontally and is retracted. It also appears smaller than normal vertically (Figure 2).



Figure 2A and 2B: Patient have Sprengel's deformity.

The glenoid appears to point downward because the inferior angle is rotated medially. A characteristic lump can be seen in the suprascapular region because the superior medial angle of the scapula is rotated upward, giving the ipsilateral side of the neck a fuller appearance. The inferior scapula is rotated more medially (Figure 3).

I15.5 degrees

Figure 3: Patient have Sprengel's deformity and limiting abduction 115.5°.

There is usually an omovertebral connection that is trapezoidal or fusiform in shape. The omovertebral connection exists in about one-third of cases [21,22]. It is a trapezoidal or fusiform structure usually located in a strong fascial capsule, extending from the superior medial angle of the scapula to the spinous, lamellar, or transverse processes of the cervical vertebrae (most commonly the fourth to seventh cervical vertebrae). It may be fibrous, cartilaginous, or bony.

The omovertebral connection is usually unilateral. It is the main cause of limited shoulder motion in patients with Sprengel's deformity. It always involves a fixed, elevated scapula and plays a major role in determining the shape and malposition of the scapula. The scapular spinae muscles are also adversely affected. The trapezius, levator scapulae, pectoralis major, rhomboids, serratus anterior, latissimus dorsi, or sternocleidomastoid muscles may be absent, hypoplastic, or contain numerous fibrous adhesions.

The trapezius is the most commonly affected muscle. Winging of the scapula may occur if the serratus anterior is weak. Sprengel deformity is usually painless. On examination, passive range of motion of the shoulder joint, including initial abduction, internal rotation, and external rotation, may be normal. Typically, thoracic scapular motion is limited. The most common limitations are in abduction and forward flexion. Abduction is limited to less than one hundred degrees in 40% of patients, which is more likely if vertebral osteophytes are present. Clinically, scapular fixation causes limited shoulder elevation and this is due to three factors:

- Vertebral bone.
- Medial border adjacent to the spinous process of the adjacent vertebra.
- The superior angle of the scapula is curved forward over the top of the chest.

The shoulder and both upper and lower limbs, spine and limbs are examined by noting scapular prominence, scapular curvature, scoliosis, thoracic asymmetry or any associated morphological abnormalities. All patients are examined for deformity. Forward flexion and abduction of the shoulder are examined, as well as range of motion, particularly scapulothoracic motion, to determine whether the scapula is anchored to the spine.

Preoperative findings were recorded including detailed patient history, findings on examination, family history of similar problems, other skeletal abnormalities such as scoliosis, omovertebral bones, Klippel-Feil syndrome, congenital vertebral abnormalities such as anterior chest wall defects, rib abnormalities and any problems during pregnancy were recorded. Indications for surgery were divided into one of the following 3 categories:

- 1. Severe limitation of the range of shoulder abduction,
- 2. Interference with activities of daily living such as difficulty in dressing oneself or
- 3. Cosmetic deformity, or both.

Associated musculoskeletal abnormalities such as Omovertebral bones, Klippel-Feil syndrome, scoliosis, spina bifida and rib abnormalities were also recorded.

The Cavendish classification was used to assess cosmetic appearance. Range of motion, pediatric outcomes data collection instrument (PODCI), and simple shoulder test (SST) were performed to assess shoulder function. Scapular position and degenerative disease were assessed using radiographs and Rigault classification.

Associated Abnormalities

Sprengel deformity almost never occurs as an isolated anomaly. It is usually associated with other anomalies, especially of the cervicothoracic spine or rib cage. The most common associated defects are scoliosis, Klippel-Feil syndrome or fused ribs, chest wall asymmetry, spina bifida or cervical ribs. The most common anomalies are hypoplasia or absence of the pectoralis major, rhomboids, trapezius, latissimus dorsi, and serratus anterior muscles. Eulenberg and Greig considered shortening of the levator scapulae to be an important etiological factor because it can be present in most cases at any severity. Syndromes associated with this condition include Greig syndrome [28] (characterized by polydactyly, syndactyly, hypertelorism, large head, and high, protruding forehead), Poland syndrome [29] (characterized by hypoplasia or absence of the pectoral muscles on one side of the body and syndactyly of the hand on the same side), Klippel-Feil syndrome, and VATER syndrome [30] (characterized by vertebral defects, imperforate anus, tracheoesophageal fistula, radial dysplasia, and renal dysplasia). The most common syndrome is Klippel-Feil syndrome. Other rare syndromes include DiGeorge syndrome [31] (characterized by heart defects, cleft palate, autism, learning disabilities, recurrent infections, and hypocalcemia), Floating-harbor syndrome (characterized by short stature, skeletal growth retardation, delayed communication, and distinctive facial features), Goldenhar syndrome (characterized by incomplete development of the ears, nose, soft palate, lips, and mandible). X-linked hydrocephalus syndrome and psychiatric disorders. The relationship between diastematomyelia (a condition in which part of the spinal cord is detached, usually at the level of the upper lumbar vertebrae) and Sprengel deformity has also been described.

Classification

The Cavendish classification is widely used and recommended for cosmetic classification of Sprengel deformity.

Grade 1 is the mildest, when the shoulder is almost flat and not noticeable when wearing clothes. Grade 2 is also mild, but the medial superior portion of the scapula is visible as a lump. In grade 3, the deformity is moderate, visible, and the affected shoulder is two to five centimeters higher than normal. In Grade 4, the deformity is severe, with the scapula being very high, with the medial superior angle at the occiput, with cervical webs, and a short scapula. A limitation of this classification is that it is difficult to apply in bilateral cases.

Clinical findings

The patient and parents note a deformity of the upper back with limited mobility of the ipsilateral shoulder. The deformity is present from birth, but cosmetic asymmetry and functional impairment are rarely noted before the age of 1 year. Most commonly, the deformity is mild and unilateral, but bilateral cases have been reported. The Cavendish classification is used to describe the clinical findings (Table 1). The deformity affects males and females equally. This malformation involves the omovertebral connection, which can be a fibrous or bony connection between the cervical spine and the scapula, occurring in 20 - 25% of cases (Cavendish, 1972). Cosmetic deformity and loss of shoulder function are common patient complaints.

Grade	Vertebral level	
Grade 1 - Very mild	Shoulder joints are level.	
	Deformity is invisible when patient is dressed.	
Grade 2 - Mild	Lump in the web of the neck.	
	Deformity is visible when dressed.	
Grade 3 - Moderate	Shoulder elevation 2-5 cm.	
	Deformity is easily visible.	
Grade 4 - Severe	Superior angle of the scapula is near the occiput with/without	
	neck webbing or brevicollis	

Table 1: Cavendish classification.

Imaging

Imaging studies

Routine chest and cervical spine radiographs should be completed as part of the evaluation. Rigault's classification is used to describe radiographic findings. In addition, radiographs should be evaluated for omovertebral bone, cervical spine abnormalities, and rib deformities (Table 2). CT, including three-dimensional reconstruction (Yamada 2013), can provide information about the omovertebral bone and scapular position. MRI is more useful in determining whether there is a fibrotic band of omovertebral tissue.

Radiographic Rigault's Classification		
Grade 1	Superomedial angle lower than T2 but above T4 transverse process	
Grade 1	Superomedial angle located between C5 and T2 transverse process	
Grade 3	ade 3 Superomedial angle above C5 transverse process	

Table 2: Radiographic Rigault's classification.

On imaging studies, the omovertebral bone is sometimes difficult to recognize due to the possibility of overlying bone.

Anteroposterior radiographs of the chest and both shoulders are the best way to identify Sprengel deformity. Leibovic., *et al.* described a method for calculating the rate of scapular displacement, using three lines drawn on the anteroposterior radiograph, which calculate the superior scapular angle and inferior scapular angle, allowing the viewer to visualize the rotation of the scapula [32]. CT scans may be performed to visualize the affected area, delineate the attachment points of the omovertebral bone, or to determine the presence of spina bifida occulta or intraspinous lesions prior to surgery [33]. Appropriate imaging studies should also be performed for any associated abnormalities.

Outcome scores

Aesthetic appearance, radiographic improvement, and functional outcomes of the shoulder at follow-up were evaluated in patients with Sprengel deformity who underwent a modified Woodward procedure. The Cavendish score and the radiographic Rigault score were used to assess the severity of the deformity and the position of the scapula relative to the cervical spine before and after surgery. Functional and subjective assessments were evaluated using the PODCI (Pediatric Outcomes Data Collection Tool) and the Simple Shoulder Test (SST). The Cavendish classification was used to assess aesthetic appearance.

Cavendish classification:

Patients with Sprengel deformity often present with impaired aesthetics and function. Consistent with the current literature, we used the Cavendish classification [16] to objectively evaluate surgical treatment.

Overview of Sprengel Deformity in Children

This classification is easy to use and more accurately assesses the severity of deformity and improvement after surgery. Cavendish in a series of one hundred cases from a series of hospitals in the UK found moderate to severe malformations and mild malformations in 59 cases. Diagnostic errors can occur even though the main features of the condition are known. Klippel-Feil syndrome, Erb's palsy and Scoliosis are some of the conditions that are difficult to diagnose. They can be differentiated quite simply with proper clinical examination and radiography. Children with Sprengel malformations have reduced range of motion (ROM) and varying degrees of functional limitations due to the shoulder joint. Lack of thoracic motion and downward rotation of the glenoid result in reduced shoulder extension.

Shoulder extension is usually limited to 90°. These limitations affect many activities of daily living. It is not easy to classify malformations when the malformations are bilateral and of similar severity.

To simplify treatment indications and describe outcomes, Cavendish proposed the following grades in 1972 [21] (Table 1): Grade 1 -Very mild, the shoulder joint is level, the deformity is not visible when the patient is dressed. Grade 2 - Mild There is a mass in the cervical membrane and the deformity is visible when dressed. Grade - 3 will have a shoulder elevation of about 2 - 5 cm and the deformity is easily visible. Grade -4 is severe and the shoulder is so high that the upper angle of the scapula is close to the occiput, with or without cervical ligaments or a shortened scapula.

The Cavendish grade is used as a measure of cosmetic improvement and the range of extension in the shoulder as a measure of function [21].

Scoliosis was found to be the most common associated abnormality in two large series of 112 patients and 75 patients (35% and 55%, respectively). Rib malformations are also common, with a reported incidence of 16% to 48% in patients with Sprengel malformation. Other associated disorders include diastematomyelia.

Rigault score

Any patient with Sprengel deformity should undergo radiographic evaluation as an initial assessment. The level of the scapula relative to the vertebrae and the contralateral side should be assessed first. It is also useful to determine the presence of associated abnormalities (e.g. scoliosis, rib abnormalities, cervical vertebrae).

The Rigault classification described by Rigault., *et al.* is based on the projection of the supermedial angle on radiographs [18,34]. It is used to assess the level of the scapula relative to the vertebrae on preoperative and postoperative radiographs. It is a simple, easy-to-calculate, and widely used scoring system. This scoring system does not correlate with the Cavendish score.

Radiographic evaluation includes measuring the ratio of superior displacement and the height-to-width ratio of the scapula in the posterior scapular view from preoperative images such as comparing the chest radiograph with the postoperative images. Scapular position and degenerative disease are assessed by radiographs. Broca's "scapular index" describes the relationship between the longitudinal length and transverse width of the scapula. This index is expressed as (100*width/length) and is calculated to be approximately 63-71 [14].

An accurate method for identifying Sprengel deformity is a chest X-ray, sometimes called an anteroposterior chest X-ray. The term "anteroposterior" means "front to back". The patient is placed on the film and the X-ray is taken from the front during this examination. The affected scapula will be elevated and rotated, with the inferior angle pointing laterally.

Plain X-rays can be used to assess the deformity and the presence of scapular-vertebral interface, and to document post-operative correction. The distance between the inferior and superior medial angles of the scapula and the spine, and the glenoid tilt angle, for assessing postoperative outcomes have been described by Ahmad., *et al.* [35] but have not been widely used. Based on the frontal radiograph and the relationship between the supermedial angle and the spine, Rigault proposed a classification to assess the deformity between the supermedial angle of the scapula and the spine.

Rigault., *et al.* developed a radiographic classification for Sprengel deformity (Figure 4) based on the radiographic projection of the supermedial angle of the scapula. The deformity is classified based on the relationship between the supermedial angle of the scapula and the associated vertebral level. Grade 1 deformity involves the supermedial angle below T1, grade 2 deformity is between T1 and C5, and grade 3 deformity is above C5.



Another classification described by Ross and Cruess is the measurement of shoulder height based on the level of the center of the humeral head relative to the vertical axis of the trunk. Leibovic., *et al.* described the Sprengel deformity as having a rotational component and measured the scapular displacement based on the vertical position of the scapula and the rotational component, assuming that the center of rotation of the scapula passes through the clavicular joint [32].

To detect scapular position, (early) degenerative changes and anatomical differences between both shoulders, standard radiographs of the anteroposterior angle of both shoulders with a chest radiograph were performed. In patients with associated abnormalities, a 'Displacement Ratio' measurement was performed by drawing two reference lines.

Line 1 was drawn from the center of the glenoid cavity of the affected shoulder perpendicular to the vertebral axis and line 2 was drawn from the normal shoulder. The above displacement ratio is defined as the distance between these two lines as a fraction of the scapular height on the normal side [33]. This ratio was not measured in bilateral cases because there was no normal side to use as a reference.

The radiographic assessment of the height-to-width ratio is the height of the scapula measured from the superior to the inferior angle parallel to the glenoid cavity, and the width of the scapula measured from the glenoid cavity to the most medial portion of the vertebral contour perpendicular to the glenoid cavity. To assess postoperative scapular hypoplasia, the inferior medial angle of the scapula was used as the reference point [24]. These measurements were assessed from preoperative and postoperative radiographs. Objective measurement of scapular hypoplasia is difficult and controversial due to the hypoplastic nature of the affected scapula. Associated spinal deformities, scoliosis, and hemivertebrae have been noted (Figure 5).

Standard anteroposterior view of both shoulders or chest X-rays were performed, with a few requiring cervical, thoracic and lumbar spine X-rays in patients with associated abnormalities. Measurement of the above displacement ratio was performed by drawing two reference lines.



Figure 5: Elevating scapula in roentgenography.

To assess the height-to-width ratio, the height of the scapula was measured from the superior to the inferior angle parallel to the glenoid cavity, and the width was measured from the glenoid cavity to the most medial part of the vertebral contour perpendicular to the glenoid cavity on the radiograph. To measure postoperative scapular hypoplasia, the medial inferior angle of the scapula was used as the reference point for assessment. Associated spinal deformities, scoliosis, and hemivertebrae were also recorded postoperatively.

The Cavendish score and the Rigault radiographic score were used to assess the severity of the deformity and the position of the scapula relative to the cervical spine, respectively, and the functional score was also assessed using the pediatric outcomes data collection tool (PODCI) and the simple shoulder test (SST).

CT scan

CT scan with 3D reconstruction was used to assess the scapular connection, scapular dysplasia, and malalignment. It is beneficial for preoperative planning.

Computed tomography (CT) scans are two sophisticated imaging modalities commonly used to confirm the diagnosis of Sprengel's malformation. X-rays and computers are used during a CT scan to create films that show cross-sectional images of certain tissue features.

Magnetic resonance imaging (MRI)

Magnetic waves are used during an MRI to create cross-sectional images of body tissues. These tests are capable of detecting vertebral bones. The use of advanced imaging techniques can also help plan and monitor surgical procedures.

Differential diagnosis

Possible differential diagnoses at presentation include:

- Rickets
- Osteoporosis
- Paralysis (especially of the long thoracic nerve)
- Nonunion of the scapula.

Management

Non-surgical management includes exercises, passive stretching, and attempts to conceal the deformity by padding the opposite shoulder. However, no significant improvement has been achieved with these methods.

The most common indication for surgery is primarily cosmetic, and loss of abduction is also an important functional loss. The ideal time for surgery is between 3 and 8 years of age, although it can certainly be performed after this age, with poor results. Indications with severe cosmetic concerns or functional deformities (abduction < 110-120 degrees).

The following factors must be considered when selecting patients for surgery:

- Unilateral or bilateral deformity
- Functional impairment
- Degree of cosmetic appearance
- Age of the patient
- Other associated abnormalities that may mask the shoulder deformity or may not provide significant results.

The literature describes various surgical treatments. These are:

- 1. Putti
- 2. Delchef (1922)
- 3. Schrock (1926)
- 4. McFarland (1950)
- 5. Green (1957)
- 6. Allan (1964)
- 7. Woodward (1961).

Initially, Putti's method of lower scapular implantation was by dividing the muscle attachments and cutting off the protruding upper part of the scapula. Based on the above technique, various modifications to bring the scapula down have been described by Schrock, Green, Mears and Woodwards [21].

Number of Patients	BCS scores	Severity of depression
1	8	Mild
2	10	Mild
3	10	Mild
4	14	Mild
5	14	Mild
6	14	Mild
7	14	Mild
8	15	Mild
9	25	Sever
10	26	Sever
Total	150	

Table 3: Scores of HAMD in Breast Conserving Surgery group. Significance of differences p<0.000232.

The scapular fusion methods of Putti (1908), Schrock (1926), and Allan (1964) were generally disappointing. Often, the deformity recurred due to the scapula returning to its previous position or due to bone remodeling after subperiosteal resection.

Several surgical techniques have been developed and evaluated for this purpose, which can be classified as partial scapulothoracic resection or scapulothoracic osteotomy [25,36]. The scapula is displaced by replacing the attachments of the scapular girdle muscles on the scapula or by replacing the scapular girdle muscles from their origins on the vertebrae. These procedures are performed as a single procedure or in combination with clavicle osteotomy to prevent brachial plexus injury.

One of the third types of surgical procedures was introduced by Woodward in 1961 [1]. Over the years, several modifications of this technique have been reported. Various procedures including the Putti and Green procedures, Mears procedures, and modified Woodward procedures have been used to correct the deformity. Various surgical procedures have been described for scapular elevation, all of which have shown reasonable and comparable results. In the original Green procedure, the muscle resection was performed distally rather than proximally. This procedure required extensive dissection and was technically demanding [37,38]. The modified Green procedure used radiographic geometry to quantify scapular depression and rotation, but the depression did not change significantly over time with recurrence of malrotation after 2 years.

Green procedure

The Green procedure is an extensive procedure that involves extensive resection and is also technically demanding [39]. Leibovic and Belleman have made numerous modifications to this procedure, to name just a few. All muscles are released extraperiosteally from their attachments. The trapezius is reflected from its medial attachment to expose the Latissimus dorsi, serratus anterior, levator scapulae, supraspinatus and rhomboids. The supraspinatus fossa is then carefully dissected to avoid the neurovascular bundle and the vertebral bones, if present, are also dissected. Once the scapula has achieved its new position, the muscles are reattached and muscle lengthening is performed to maintain the new position. Once the final attachment points are made, the shoulder joint should be stable and not displaced medially. Velpeau tape is applied for two weeks and the child is then encouraged to perform swing exercises for several weeks before starting active range of motion exercises.

Modifications of the Green's technique

Leibotic he improved the Green technique by anchoring the scapula into a pocket created in the Latissimus dorsi muscle [32].

Bellemans and Lamoureux

They avoided releasing the Serratus Anterior muscle to allow for immediate postoperative mobilization. Andrault advocated clavicle resection to prevent brachial plexus palsy. The main drawbacks of this procedure are recurrence after two years, a few cases of postoperative scapulothoracic resection, and a higher risk of brachial plexus injury in older children. The recommended upper limit for surgery is 8 years [37].

Although the modified Green method has shown good cosmetic and functional results and has encouraged more surgeons to attempt this method, it has its own drawbacks. There is a higher risk of keloid formation and postoperative scarring. As mentioned earlier, the Green procedure has a steep learning curve and involves extensive dissection and requires solid technical skills.

In contrast, the Woodward procedure has a lower incidence of hypertrophic scarring and provides much better results and has been adopted as the gold standard. This has been cited in several studies and has produced excellent results. The Woodward procedure is the gold standard with over 80% successful cosmetic and functional outcomes.

Mear technique

In 2001, Dana Mears described a new surgical technique that included a scapulothoracic osteotomy, partial scapulothoracic resection, and release of the long head of the triceps. This technique involves an oblique osteotomy of the supraspinatus fossa to adequately resect the scapula to prevent impingement [6]. The long head of the triceps and part of the origin of the Teres minor muscle are released to increase the range of extension and the scapulothoracic resection is in an inferomedial direction to allow 160° of shoulder extension.

Clavicle resection may be considered depending on the extent of brachial plexus involvement. Results from some studies have shown a gradual improvement in range of motion. Recent reports have shown problems with postoperative keloid formation, delayed postoperative mobilization until the wound heals, and less scapular lowering when compared with the modified Woodward procedure. However, this technique is relatively new and there is little literature on its application.

Woodward operation

The Woodward operation was introduced by Woodward in 1961 and hence the name [6,40] (Figure 6). The procedure involves separating the origins of the rhomboid and trapezius muscles from the spinous process, resecting the spine and fibrous band if present, and moving the scapula downward [41-44]. Postoperatively, a Velpeau sling was used to immobilize the limb and range of motion exercises were started 3 weeks postoperatively.

The modification of the Woodward operation [11,19] included the addition of a medial superior border resection and a supraspinous resection of the scapula. Approximately 1 cm of the medial border of the scapula was resected and the resection was performed superiorly and medially to the scapular notch.

After a midline incision, the vertebral attachments of the trapezius, rhomboids, and levator scapulae were separated to allow rotation and displacement of the hypoplastic left scapula more posteriorly on the chest (Figure 6).



Figure 6A and 6B: A: Drawing to show the anatomical features of congenital elevation of the scapula (trapezius cut away). B: Drawing to show excision of the supero-medial part of the scapula. Stippled areas indicated bone [43].



Figure 7A and 7B: A: Drawing to show Woodward type scapular transplantation Stippled areas show excised bone. B: Before trapezius slide and showing displacement of scapula after trapezius slide.



Figure 8A-8C: Showing Omovertebral bone intra-operation. A: Omovertebral bone; B: The Omo-vertebral part is cut off; C: Superomedial angle of scapula.

Modified Woodwards surgery

Under general anesthesia, the patient is placed in the prone position. The arm with the scapular mesh is left free for manipulation during the procedure. The entire arm, shoulder and the area from the nape of the neck to the upper occipital root to the lower lumbar region are prepared and draped. A 15 cm midline incision is made over the spinous processes extending from C4 distally to the spinous process D10 (Figure 7). The skin and subcutaneous tissue are weakened laterally to the medial border of the scapula on the affected side and the lateral border of the nerves to the arm. The origin of the trapezius muscle from the spinous processes C2 to D10 is dissected and the plane between the trapezius and serratus anterior is developed (Figure 8). The Rhomboids insertion is cut proximal to the scapula. The superior medial border of the scapula together with its Omovertebral attachment is removed. The separated trapezius muscle is sutured at a lower level, pulling the scapula down. The scapula is reduced, with the trapezius and rhomboids reattached in a more caudal positioned beneath it. The latissimus dorsi is sutured to the lower end of the scapula. During reduction, there is a possibility that the nerves of the brachial plexus may become entrapped between the chest wall and the clavicle. In older children and Rigault grade -3, a claviculectomy is performed to prevent brachial plexus injury. The scapula is reduced, with the trapezius and rhomboids reattached in a more caudal position to the interspinous ligaments in the midline (Figure 9).

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Figure 9: Modified Woodward's surgery. Excision of supero-medial portion scapula along with Omovertebral bone is removed.

Clavicular osteotomy

Make an incision approximately 2 cm above the midclavicle. Create a plane below the dermatome, incise the periosteum longitudinally and expose the clavicle under the periosteum. Perform a clavicle resection and use a rongeur to create cortical bone fragments over the midclavicle approximately 1 cm in length. A clavicle resection is required in older children or in cases of associated deformities. The wound is closed in layers over a suction drain. Sometimes a VAC dressing is applied based on surgeon preference.

At follow-up, the data are evaluated for deformity using the Cavendish classification as a measure of cosmetic improvement and the range of shoulder abduction as a measure of function. Radiographic evaluation is performed using the Rigault classification. This classification is based on the position of the superior-medial angle of the underdeveloped scapula.

Sprengel deformity is a congenital prominence of the scapula during embryonic development. In growing children, this impairment is static because the fibrous tissue does not grow [18]. This deformity can have a serious impact on activities of daily living and shoulder function [18]. Limited shoulder extension and aesthetics are major concerns. Nonsurgical treatment has not been successful in the treatment of Sprengel deformity [3,9,18,34,45]. Several surgical procedures have been described for the correction of the above deformity. Among the procedures described such as Woodward, Green, Mears and their modifications, the modified Woodward procedure has been found to be the most commonly used procedure. Although many studies have been reported, very few studies have described the functional outcomes of surgical correction of Sprengel deformity. We used the Cavendish score to assess cosmetic improvement, the Rigault score, scapula height-to-width ratio, and superior displacement ratio to assess radiographic improvement, the PODCI (Pediatric Outcomes Data Collection Instrument) score, and the Simple Shoulder Test (SST) score to assess functional improvement.

Cavendish and Rigault scores are widely used in the literature [1,8,33] and form a common basis for comparing results between studies. We used these scores because they are easy to use and because they more accurately assess the degree of deformity and improvement after surgery. Jindal N., *et al.* [17] reported on twelve children who underwent the Woodward procedure at a mean age of 5.8 years, preoperative mean.

Cavendish score was grade 3 [21] and mean shoulder abduction range was 115.83°. At a mean follow-up of 31.8 months, there was a statistically significant improvement to a Cavendish grade of 1.25 and an abduction range of 153°.

Similarly, Walstra., *et al.* [1] reported on 8 shoulders at a mean age of 14.7 years that showed cosmetic improvement from preoperative Cavendish grade 3 to grade 1 or 2 at final follow-up. In our study, the average cosmetic improvement of 2 Cavendish grades (from 3.43 to

1.42) in 14 shoulders was assessed with a significant P value <0.001 higher than that reported. One of the main reasons for this was the extensive resection of the superomedial angle, an essential component of the modified Woodward. When compared with the modified Green, the long-term results reported by Gonen., *et al.* [46] in 24 children showed an improvement of at least one Cavendish grade in 88.9% of shoulders. The results of Mear's procedure focused primarily on improvement in range of motion and cosmetic comparisons are not possible based on the available literature. Radiographic improvement of scapular demotion was assessed using the Rigault scoring system, again a commonly used outcome measure for this modality. Walstra., *et al.* [1] using the Woodward procedure showed a significant with a p-value of 0.008. The mean scapular demotion in our study was 2.01 cm, which is comparable to the value of 2.2 cm reported by Siu., *et al.* [33].

Outcome comparisons were also assessed in patients with associated deformities versus those without. Patients without associated deformities had a significant improvement in Cavendish Rigault grade with p-values of 0.046 and 0.008, respectively. All other factors between groups did not show significant values. This supports the established practice of avoiding surgery or operating with patient consultation for cosmetic improvement when there is an associated abnormality.

Range of motion is an important factor in shoulder function and has been specifically targeted in surgery as described by Mears. There was a significant improvement in abduction and forward bending in our patients. This was more significant in the forward bending range (mean improvement of 40°). This was more significant in children under seven years of age. Previous studies of the outcomes of the Woodward procedure have reported a mean functional improvement in abduction of 32-59° and few complications.

The improvement in shoulder abduction in our series is consistent with these results. Our study showed an improvement in the abduction range with a mean of 36.5° (P < 0.001) at a mean follow-up of 54 months. Other Woodward series such as Walstra., *et al.* and Mushin., *et al.* [32] also showed improved range of motion over the long term. Mears., *et al.* showed a greater abduction improvement of a mean of 60° in 8 patients compared to the Woodward procedure [40]. The Mears procedure is a very important procedure involving release of the triceps from the glenoid and osteotomy through the glenoid body and as the functional improvement has not been shown to be superior, it is controversial whether a more extensive procedure should be performed compared to the Woodward.

When we combined our data with eligible published data regarding abduction, the combined data of 14 cases confirmed significant improvements in abduction, forward flexion, scapular displacement, and height-to-width ratio postoperatively and at follow-up.

Leibovic., *et al.* [32] modified the Green technique by suturing the shortened scapula into a pocket formed in the latissimus dorsi muscle. In 16 shoulders (14 patients), with a mean follow-up of 6.5 years, the mean shoulder abduction was reported to be 148° (range 100° to 180°), compared with 91° preoperatively (range 60° to 120°). Although the initial results were promising, scapular rotation was not maintained at long-term follow-up, and half of the cases developed hypertrophic scarring.

Quality of life and functional outcomes after surgery for Sprengel deformity have been measured using scoring systems such as the Constant Score, DASH (Disability of Arm, Shoulder and Hand Score), and SST (Simple Shoulder Test). The PODCI score has previously been reported to be a valid and reliable tool for assessing function in conditions such as cerebral palsy, congenital muscular dysplasia, and unilateral upper limb disability (U-UED) when compared with a group of normal children. To our knowledge, the PODCI scoring system has never been used before to study outcomes for Sprengel deformity.

On the other hand, the SST has been used for Sprengel deformity and is considered acceptable with demonstrated good reliability and test-retest validity. Compared with four other shoulder scales such as the American Shoulder and Elbow Surgeons Standard Shoulder Assessment, Shoulder Pain and Disability Index, and SSI, the structural value of the SST has a fairly good correlation.

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Several studies evaluating Sprengel have evaluated function using the Constant Score and DASH (Disabilities of the Arm, Shoulder, and Hand Score). The Constant Score was not originally intended for pediatric evaluation, is not standardized for children, and the strength component has significant and unreliable variability across age and gender, and is not a good outcome assessment tool. To our knowledge, the PODCI score has never been used in the literature to assess functional outcomes for Sprengel deformity. The scoring system is easy to use and reliable. We chose the PODCI and SST scoring systems to evaluate functional outcomes. A mean PODCI score of 83.21 and a mean SST score of 9.7 were found in our patients. The maximum SST score was fixed at 12 points. Better shoulder function was valued higher. The mean SST score obtained from the study by Walstra EF. *et al.* [1] for patients with Sprengel anomaly corrected by the Woodward procedure was 9.5 points (range 7-12). The findings of the prospective study by Roy., *et al.* showed that the difference in clinical importance of the SST was 3 on the 12-point SST scale. Parental and patient satisfaction with the results of the modified Woodward procedure was rated as good at follow-up in all patients. The PODCI score of 83.21 was significantly higher than the standard score of 50, indicating that the function in these children was above average and not impaired.

Complications

Postoperative complications include nerve palsy, brachial plexus injury, incomplete correction, recurrence, wound infection, and surgical scar appearance. The brachial plexus is at risk of compression during surgery because of the inferior displacement of the scapula.

Some authors have reported brachial plexus palsy in 6 - 11% of patients after Sprengel malformation surgery, and this tends to occur in children with larger and more severe malformations; however, many of these palsies are temporary. Consideration should be given to clavicle resection to prevent brachial plexus injury in these groups. Intraoperative sensory evoked potential monitoring may help prevent such injuries.

In the literature, the incidence of hypertrophic scarring is 26 - 64%, recurrence of the upper pole of the scapula is 30%, and scapular wing is 4 - 17% [9]. The Green technique has been reported to cause keloid scarring due to the incision site on the shoulder, recurrence of scapular elevation after 2 years, and postoperative scapular winging in some cases.

Complications reported in previous series include open scars, scapular winging, brachial plexus injury, and long-term complications such as keloid formation and poor scarring. The brachial plexus is at risk of intraoperative compression because the scapula is pushed downward.

Postoperative complications include nerve palsy, brachial plexus injury, incomplete correction, recurrence, wound infection, and surgical scar appearance. The brachial plexus is at risk of intraoperative compression because the scapula is pushed downward.

In this study, we had one patient with brachial plexus injury and two patients with hypertrophic scars. Previous studies have reported that the best time for surgery is between 3 and 8 years of age because at older ages, clavicle resection is required to prevent brachial plexus injury and the degree of correction is less.

Compared with the Woodward technique, the modified Green technique has been reported to cause keloid scarring due to the incision site on the shoulder, scapular elevation after 2 years, and scapular winging after surgery in some cases. Regarding the risk of keloid formation and postoperative scarring, Greitemann B., *et al.* (1993) [23] reported better results when using the Woodward method than the Green method in the treatment of congenital scapular elevation in 37 cases of Sprengel deformity.

Hung and Hien 2018 [47] reported: Surgical treatment of Sprengel deformity of the scapula in children. Fourteen patients (14 shoulders) who underwent the Woodward procedure and the modified Woodward procedure for Sprengel deformity were evaluated.

Patient and parental expectations must be carefully managed. The goals of nonsurgical treatment include maximizing shoulder strength, range of motion, and function. However, nonsurgical treatment does not correct the deformity. Complications of surgical intervention include persistent deformity, hypertrophic/wide scar, scapular winging, brachial plexus injury, and recurrent deformity.

Aesthetic appearance and function: Role outcomes were evaluated using the Cavendish grading system and continuous score function. Grading of aesthetic appearance, shoulder range of motion, and radiographs were performed for interpretation.

Woodward procedure and/Modified Woodward procedure with Age at Surgery: Age at surgery: 9+1 (SD = 46.10)/6+8 (SD = 26.62); Pvaluate: 0.00000; Age at Lase Follow-up: 14+3 (SD 35.34)/11+1 (SD = 0.35); Pvaluate: 0.10928; Cavendish grade at Preoperation: 3 (SD = 0)/3.13 (SD = 0.35); Pvaluate: 0.00000; Cavendish grade at Last Follow-up: 1.16 (SD = 0.40)/1.38 (SD = 0.52): Pvaluate: 0.560689 (not significantly different); Motion (°) at Preoperation: 97.5 (SD = 8.80)/93.13 (SD = 4.58); Pvaluate: 0.005416; Motion (°) at Postoperation: 148.33 (SD = 10.32)/147.5 (SD = 6.54); Pvaluate: 0.596971 (not significantly different); Scapular Lowering on X-ray (cm): 2.53 (SD = 0.76)/2.21 (SD = 0.70); Pvaluate: 0.496 (not significantly different).

Rehabilitation following surgery

After surgery, the patient's shoulder is immobilized to promote healing and reduce any pain that may occur. Reducing deformity and allowing normal shoulder movement is the goal of physical therapy.

- TENS stands for transcutaneous electrical nerve stimulation.
- Transcutaneous electrical nerve stimulation, which can be activated by stretching exercises, helps reduce pain.
- Ultrasound: Ultrasound can easily break up adhesions. Thermodynamic therapy: Using heat therapy, the muscles are relaxed before starting an activity program.
- Mobilization: Six weeks later, passive and active range of motion exercises will be the focus of physical therapy. Progressive relaxation
 and passive movement of the shoulder and scapula are performed to increase range of motion and flexibility of the shoulder. Early
 mobilization of the shoulder and scapula, including abduction and elevation.

Strengthening exercises

All exercises that target specific muscle groups help strengthen weaker muscles. The shoulder girdle muscles can be strengthened with isotonic and isotonic exercises including bridges, arm supports, forearm supports, shrugs, and push-ups.

Upper trapezius stretch

The child sits in a comfortable chair. The child places his right hand directly under his groin. The therapist supports the child's shoulder by holding the lateral border and shoulder blade. The child then flexes his neck and rotates his neck to the left. And with that movement, the child places his left hand directly under his knee. The therapist supports the child's shoulder by holding the lateral border and shoulder blade. The child then flexes his neck and rotates his neck to the right. The child holds this neck position for ten seconds while the stretch is performed.

Shoulder blade stretch

The child lies on his back. The right shoulder is flexed ninety degrees and the elbow is fully extended. The infant then uses all of his strength to push the right elbow forward. The therapist holds the shoulder blade and grasps the scapula to prevent the infant's scapula from rising and the trunk from rotating.

Posterior tilt exercise for the scapula

With the humerus above, the shoulder extended 130° to 145°, and the forearm neutral, the infant lies on his back. The infant then gently places the back of the left hand on the forehead, below the eyebrows. As the patient extends the right arm with the elbow extended, the therapist prevents the infant from raising the scapula.

Family education

Parents should supervise their child throughout each exercise session. When exercising at home, parents should keep an eye on their child to maintain proper shoulder blade mobility and prevent the shoulder blade from rising. If the upper edge of the shoulder blade slides upward, you should stop the activity and have your child try again.

Prognosis

For people with Sprengel's shoulder, the prognosis can vary depending on the severity of the problem and whether treatment is effective. Early diagnosis and treatment can lead to better outcomes and improved shoulder function. It is important to remember that Sprengel's shoulder is a complex condition and each case will present and respond differently to treatment. You should be evaluated and treated by a trained health care professional if you believe you or someone you know is affected by this condition.

Illustration

A girl, age at time operation was 4 years, 6 months; age at last follow-up was 9 years, 2 months (Postoperative 4 years and 8 months).



Figures 10A-10F: A: Preoperative with Cavendish grade 3; B: Abduction of the Shoulder was 95°; C: Roentgenography shown high

left Scapula. D: Postoperative 3 weeks, Cavendish grade 1. E: Cavendish grade 1 at last follow-up; F: Abduction of the Shoulder was

140° at last follow-up.

Currently, the preferred method is the modified Woodward procedure because it does not require a postoperative spica cast to anchor the sutures, has a lower risk of bleeding and brachial plexus injury, and has shown good results in the past. This procedure has advantages over the Putti-Chrock Green scapular bone graft and is less likely to cause extensive scarring. Because the incision is in the midline, the possibility of postoperative scapular fixation with a scar is less likely because of less dissection around the scapula and the possibility of recurrence of the deformity is also less.

Conclusion

Sprengel deformity of the shoulder is a dysplasia of the pectoral girdle, resulting in cosmetic and functional disabilities. This deformity is associated with other congenital anomalies, which often determine the management and outcome of treatment. The surgical techniques described in the literature provide favorable cosmetic and functional results, and have low complication rates.

We have demonstrated significant improvement in aesthetics, radiographic range of motion, and shoulder blade position and function using the modified Woodward procedure. The results are consistent with published literature. It is a safe and predictable procedure with very few complications.

One shoulder blade is abnormally prominent in the back in a rare congenital condition called Sprengel's shoulder. This condition can lead to asymmetry, limited shoulder motion, and associated abnormalities. Diagnosis is based on a combination of imaging, physical examination, and medical history. The goal of physical therapy is to improve posture, range of motion, and shoulder function. In more severe cases, surgery may be recommended. Early diagnosis and care are important for better outcomes.

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