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Systematic Review

Shoulder arthrodesis in the pediatric and adolescent population: A systematic review

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ABSTRACT

This study aimed to summarize the available evidence on the role of arthrodesis in pediatric patients and describe the various techniques used, their functional outcomes, and complications. AMED, CINAHL, EMBASE, MEDLINE, and Google Scholar were searched for eligible articles published until December 20th, 2022. Study screening, selection, and data extraction were performed in duplicate and independently. Qualitative synthesis was used to describe the study design, setting, aims, inclusion and exclusion criteria, patients' demographics, as well as relevant details on the surgical intervention and recovery. This review included nine studies, with one cross-sectional and eight case-series studies. The total number of included patients was 252, with a mean [standard deviation (SD)] age of 16.67 (10.3) years. The fixation method used in shoulder arthrodesis was primarily screws in 157 patients (62.3%), followed by Steinmann Pin in 61 patients (24%) and plate fixation in 35 patients (14%). The mean (SD) duration of follow-up was 6.8 (4) years, with a reported mean (SD) pooled success rate of 87.6% (9.17%), with most studies reporting an improved functional outcome. Non-union was the most reported complication in 31 (12.3%) patients. Arthrodesis is still a rather uncommon treatment used in rare situations to enhance shoulder function in pediatric and teenage patients, as alternative reconstructive techniques would likely be ineffective. Although the reported evidence on the efficacy and safety of shoulder arthrodesis is widely heterogeneous, a positive outcome was generally reported by studies regarding functional outcomes and complications.

Keywords: Arthrodesis, Orthopedic, Paralysis, Pediatric, Shoulder fusion

INTRODUCTION

Shoulder arthrodesis, also known as humeroscapular arthrodesis, is the fusion of the humeral head and glenoid. It is generally considered a salvage procedure for managing upper-extremity paralysis resulting from conditions such as poliomyelitis, tuberculosis (via spinal involvement), or rheumatoid arthritis (due to joint deformities and nerve compression), among others.[1] This procedure is generally discouraged in children due to the risk of deformity caused by epiphyseal plate injury and subsequent growth disturbance at the proximal humeral physis. [2]

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It is recommended to delay this procedure in children until skeletal maturity to avoid the risk of upper-limb motor impairment. Shoulder arthrodesis performed after skeletal maturation typically yields better functional outcomes, particularly for elbow and hand movements. In addition, instituting early rehabilitation to maximize the use of extremities is crucial.[2]

Arthrodesis is performed to fuse the glenohumeral and acromiohumeral joints in cases of severe shoulder dysfunction, particularly in conditions such as rotator cuff paralysis, brachial plexus injury, or other pathologies where tendon transfer is not an appropriate treatment option.[3] Following arthrodesis, appropriate shoulder positioning is critical. Prior evidence revealed the optimal positioning in arthrodesis with 20-55° abduction, 15-30° forward flexion, and 15-40° internal rotation.[4]

In infants, arthrodesis may be necessary to provide shoulder stability and improve function, but it is challenging to achieve because the humeral head contains a greater proportion of cartilage. A study of ten children who underwent shoulder arthrodesis for conditions such as brachial plexus injury or severe shoulder paralysis demonstrated positive outcomes, including pain relief and improved upper limb function. Follow-up evaluations indicated successful fusion and enhanced quality of life.^[5] In conditions such as brachial plexus injury or rotator cuff tear, there may be involvement of muscle atrophy, including the deltoid, infraspinatus, and supraspinatus muscles. This muscle wasting can contribute to shoulder dysfunction and is often a key factor in determining the need for surgical intervention, such as tendon transfers or shoulder arthrodesis. Furthermore, pain and motor functioning of the trapezius, serratus anterior, levator scapulae, and rhomboid muscles are assessed before performing the operative treatment to help maximize functional ability. Radiography and, in severe cases, tomography is recommended to assess internal pathologies of the shoulder, including abnormalities in the muscles, bones, and soft tissues, as well as to evaluate any associated complications such as fractures, tendon tears, or joint degeneration.[6]

Fixative techniques for shoulder stabilization may include options such as spica casting and the use of compression screws, with or without plates. These methods are considered when more conventional approaches, such as tendon transfers, are not feasible.^[7] External fixation plays a significant role in compression arthrodesis, and some researchers also advocate for both external and internal fixators. A primary advantage of external fixation is that it helps prevent the wound complications associated with plating.[1]

Thus, shoulder arthrodesis or humeroscapular is an orthopedic surgery of the upper limb due to injury or destruction in the humeral joints. It is not preferred to perform in children due to immature skeletal development

and hinders motor function development. The indications for shoulder arthrodesis include conditions such as brachial plexus injury, severe deltoid muscle paralysis (often following nerve damage), irreparable rotator cuff tears, chronic shoulder infections (such as osteomyelitis), and significant bone damage (due to trauma or degenerative disease) that results in joint instability or dysfunction. This systematic review aims to summarize the evidence on the use of shoulder arthrodesis in pediatric/adolescent patients, focusing on the variety of surgical techniques used, their functional outcomes, and associated complications.

MATERIALS AND METHODS

Literature search

A literature search of all related articles was conducted using several available databases, including AMED, CINAHL EMBASE, MEDLINE, as well as Google Scholar. The search strategy included the following key terms: "arthrodesis" OR "shoulder fusion" OR "acromiohumeral" OR "glenohumeral" OR "shoulder fixation" OR "restriction of movement" OR "joint pain" OR "paralysis" AND "pediatric" AND "children" AND "teenagers." This review was not limited to a continent, region or country of origin, and/or publication date. In this review, we included studies investigating children aged 17 years or younger who were diagnosed with a pathology requiring surgical shoulder arthrodesis procedures. Eligible study designs were clinical trials, case series, or cross-sectional studies, and the publication had to be in English. We excluded studies in which adult patients constituted the majority (over 50% of participants), as well as review articles, cadaveric studies, and animal studies, given they do not provide primary data.

Furthermore, we excluded studies focusing solely on surgical techniques without reporting clinical outcomes. All listed references within the included articles were screened for possible inclusion. The last search was done on the 20th of December 2022.

Study selection and eligibility

The inclusion criteria were studies that included children aged 17 years or under-diagnosed with pathology requiring surgical arthrodesis procedures. All studies with adult patients or with the majority of adult patients who have had the same medical procedure were excluded. Study designs considered for inclusion included clinical trials, case series, and cross-sectional studies, while review articles, cadaveric, and animal studies were excluded. Moreover, studies reporting procedure techniques without outcomes were also excluded, and studies of all levels of evidence were considered for inclusion. Two independent authors screened the titles and abstracts of potentially eligible studies and subsequently performed full-text screening of studies that

met the inclusion criteria. Disagreement between authors regarding study selection and eligibility was resolved through consensus. Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart shows the process of study selection and inclusion [Figure 1].[8]

Data extraction

Two authors (AA and WK) evaluated each of the included studies. They extracted descriptive information on the study design, setting, aims, inclusion and exclusion criteria, patients' demographics, as well as relevant details on the surgical intervention and recovery. Assessed surgical parameters included the method and technique of the procedure, angle of positioning at each position, functional outcome, and complications.

Quality assessment or risk of bias

The included studies were critically assessed using Joanna Briggs Institute critical appraisal tools for qualitative and quantitative studies.^[9] This checklist was prepared to methodically assess case-series and cross-sectional study designs; including ten criteria elements: (1) Precision of inclusion, (2) reliability of measured condition, (3) method validity, (4) consecutive patients' inclusion, (5) completeness of inclusion, (6) patients' demographics, (7) comprehensiveness of clinical diagnostics, (8) clearly reported outcomes of interest, (9) clear definition of site or clinics, and (10) the appropriateness of statistical analyses. Two independent coauthors (MA and MAA) assessed the included studies and assigned a level of quality for each article based on the following classification: poor (<60%), fair (60-89%), and good $(\ge 90\%)$.

Results of the qualitative synthesis were expressed in a narrative summary manner, with descriptive statistics, including means and standard deviations, being reported for continuous variables, while frequencies and percentages were expressed for categorical variables. The analysis was carried out using Microsoft Excel software.

RESULTS

Characteristics of the studies and participants

3341 records were retrieved from the databases, 9 of which met the inclusion criteria and were included in the qualitative

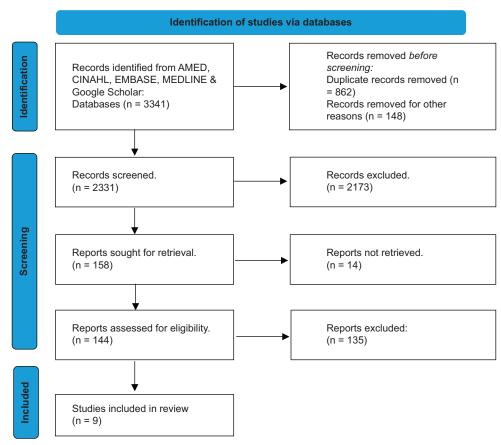


Figure 1: Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart showing the database searching process, study screening, and inclusion.

synthesis. [2,4,5,10-15] Studies were excluded because they included non-pediatric patients solely, were published in a language other than English, and utilized procedures besides arthrodesis. The included studies were published between 1942 and 2011 in 6 different journals [Figure 1].[8]

Eight studies were case series, and one was cross-sectional.[10] Three studies were conducted in the US, and the others were conducted in Canada, Germany, Hong Kong, Israel, Mexico, and the UK. The characteristics of the included studies are summarized in Table 1. The total number of study participants was 252, with a mean (standard deviation [SD]) age of 16.76 (10.3) years. Male participants represented the highest percentage (69%), with 137 participants, while two studies did not report sex.[10,13] Only two studies reported the lateralization of shoulder arthrodesis (28 right and 28 left). [4,14]

Quality assessment

The quality of the included studies was assessed based on the checklist proposed by Joanna Briggs Institute critical appraisal tools for qualitative and quantitative studies.[9] Overall, all the included studies were deemed "Fair" or above in the evaluation. Five studies (55.5%) were assessed as "Fair," [5,11-14] whereas the remaining four (45.5%) studies were deemed of "Good" quality.[2,4,10,15]

Operative technique

The percentages of the fixation method used in shoulder arthrodesis were screws in 157 patients (62.3%), followed by Steinmann Pin in 61 patients (24%), and plate fixation in 35 patients (14%). All studies reported using Spica for post-operative immobilization, with two studies^[4,14] also using abduction splints and slings for 30 participants. The average (range) degrees of fusion position for abduction, flexion, and rotation (mostly internal) were 53° (38.5-90°), 25.5° (10–40°), and 31° (20–46°), respectively [Table 2].

Clinical outcome

The mean (SD) follow-up following shoulder arthrodesis for the included studies was 6.8 (4) years. The mean (SD) success rate of fusion in shoulder arthrodesis among the participants was 87.6% (9.17%). However, one study did not provide information about the fusion success rate.[12] In terms of complications related to shoulder arthrodesis, 31 participants had non-union, 7 had an infection, 5 had pseudarthrosis, 4 had a fracture, 2 had malunion, and 1 had a hematoma. Three studies did not report complications. [5,12,13] Most studies rated the function of individuals after shoulder arthrodesis as excellent and good, reported in 65 (30.1%) and 23 (10.6%) patients, respectively. [2,5,10-15] Only one study by Miller reported that seven patients (53.8%) were able to reach their mouth.^[4] Most participants did not suffer from pain after the operation [Table 3].

DISCUSSION

Arthrodesis remains a rare but reliable method for improving shoulder function in pediatric and adolescent patients in selected cases as a secondary salvage procedure in which other reconstructive methods would probably provide little or no benefits. It can be considered inevitable for restoration of glenohumeral joint stability, provision of stability in the flail shoulder following damage to the brachial plexus, resection of malignant bone tumor in the shoulder joint, and infection-induced joint destruction.[3]

Makin advocated that the arthrodesis should be done when the child is between the ages of 5 and 9 years, as he found that children with flail shoulders will never achieve functional

Author, year	Quality	Country	Sample size	Age in years (min - max)	Sex, Female (%)	Laterality (%)
Miller <i>et al.</i> , 2011 ^[4]	Good	US	13	14.7 (7–17.8)	6 (46)	•Right: 8 (62) •Left: 5 (38)
Barr <i>et al.</i> , 1942 ^[10]	Good	US	102	12.5 (6-30)		
Kalamchi, 1978 ^[12]	Fair	Hong Kong	10	15.3	2 (20)	
Rountree and Rockwood, 1959 ^[13]	Fair	US	14	7.4 (5–15)		
Mah and Hall, 1990 ^[5]	Fair	US and Canada	10	12.75 (8–18)	4 (40)	
De Velasco Polo and Monterrubio, 1973 ^[15]	Good	Mexico	31	12.6 (8–15)	13 (42)	
Charnley and Houston, 1964[11]	Fair	UK	23	33.2 (6-58)	5 (21.7)	
Makin, 1977 ^[2]	Good	Israel	7	6.6 (5-9)	3 (42.8)	
Rühmann <i>et al.</i> , 2005 ^[14]	Fair	Germany	43	35 (11–82)	10 (23)	•Right: 20 (47) •Left: 23 (53)

Table 2: Summary of modes of intervention used in shoulder arthrodesis in included studies.									
Intervention	Fix	ation Type		Position of fusion (Average)			Postop immobilization		
Author, year	Stenman pin	Screws (%)	Plate (%)	Abduction (Degrees)	Flexion (Degrees)	Rotation (Degree)	Spica (%)	Sling (%)	Abduction splint (%)
Miller <i>et al.</i> , 2011 ^[4]	0	8 (62)	5 (38)	42.31°±18.44	23.85°±11.57	26.15 degree±11.93 internal	8 (62)	5 (38)	0
Barr et al., 1942 ^[10]	0	102 (100)	0	40	10	42 internal	102 (100)	0	0
Kalamchi, 1978 ^[12]	0	10 (100)	0	38.5	28.5	42 internal	10 (100)	0	0
Rountree and Rockwood, 1959 ^[13]	0	14 (100)	0	45–55	15–25	15–25 internal	14 (100)	0	0
Mah and Hall, 1990 ^[5]	0	10 (100)	0	45	25	25 internal	10 (100)	0	0
Velasco Polo and Monterrubio, 1973 ^[15]	31 (100)	0	0	90	25	30 internal	31 (100)	0	0
Charnley and Houston, 1964 ^[11]	23 (100)	0	0	43	42	46 internal	23 (100)	0	0
Makin, 1977 ^[2]	7 (100)	0	0	90	25	25 external	7 (100)	0	0
Rühmann et al., 2005 ^[14]	0	13 (30.2)	30 (69.8)	20-60	20-40	0-50 internal	18 (42)	0	25 (58)

Table 3: Summary of outcomes after shoulder arthrodesis reported in the included studies.							
Author, year	Follow-up years (average)	Fusion (%)	Function (%)	Pain (%)	Complication (%)		
Miller <i>et al.</i> , 2011 ^[4]	3.4	12 patient (92.3)	7 patients reaching mouth (53.8) 1 patient reaching forehead (7.6) 4 patients reaching behind heads (30.7)	None	Nonunion 1 (7.6) Malunion 2 (15.3)		
Barr et al., 1942 ^[10]	More than 1 year	81 (77.5)	excellent 10 Majority sup optimum		Nonunion 23 (22.5)		
Kalamchi, 1978 ^[12]	3.5		4 excellent (40) 6 good (60)				
Rountree and Rockwood, 1959 ^[13]	8	12 (85)	5 excellent (36) 9 good (64)	None (0)			
Mah and Hall, 1990 ^[5]	15.1	10 (100)	8 excellent (80)		None (0)		
De Velasco Polo and Monterrubio, 1973 ^[15]	4.5	25 (80)	13 excellent (41.9) 8 good (25.8) 3 Fair (9.6) 6 poor (19.3) 1 lost follow up (3.2)		Non-union 6 (19.3)		
Charnley and Houston, 1964 ^[11]	6.6	18 (78)	No specific function reported, just generally mentioning good outcome 4 lost follow-up (17.3)		Non-union 1 (4.3) Hematoma 1 (4.3) infection 2 (8.6)		
Makin, 1977 ^[2]	Until skeletal growth	7 (100)	4 excellent (57.1)		Malunion 1 (14.2)		
Rühmann <i>et al.</i> , 2005 ^[14]	6.7	38 (88)	21 excellent (49) 5 poor (12)	2 patients (4.6)	Pseudarthrosis 5 (12) Infection 5 (12) Fracture 4 (9)		

level if shoulder arthrodesis is delayed until skeletal development is adequate. $^{[2]}$ Pruitt $et\ al.$ preferred to delay fusion until 10 years of age,[16] while Rountree and Rockwood

believed that the arthrodesis should be done after the age of 8 years. However, controversy persists as to the optimum age for the arthrodesis.[13]

Various abduction, flexion, and internal rotation postures have been recommended in various studies as the optimal positions for shoulder arthrodesis. Using moiré photography, Jonsson et al. proposed a method for determining the position of shoulder arthrodesis. It generates a topographic picture of an item using grid lighting. This approach showed the scapula location and allowed for arthrodesis. According to Jonsson et al., the optimal abduction posture, internal rotation, and forward flexion should be 20-30°.[17]

Among the options are acromiohumeral, glenohumeral, and hybrid procedures for shoulder arthrodesis. The acromiohumeral method was traditionally employed. However, combination approaches stabilizing the internal fixation are now deemed acceptable.[18] For instances with little or no bone loss for glenohumeral salvage, several publications proposed employing a reconstruction plate for shoulder arthrodesis. To prevent fractures, a substantial bone shortage should be addressed with additional treatment approaches. [19] Cofield and Briggs improved the fixation technique by including three screws, two across the humeral head and one through the acromion. [20] Another study recommended employing a pelvic reconstructive plate owing to its ease of use and low visibility.[6]

The optimal arthrodesis position within the pediatric age group is discussed extensively in the literature. Pruitt et al. concluded that the angles were not critical, but hyperabduction, which gave the worst results, and excessive flexion should be avoided. [16] Most of the suggested specific ranges of different fusion positions ranged from 20 to 55° of abduction, 15 to 30° of forward flexion, and 15 to 40° of internal rotation.[4] As reported in recent literature, high angles exceeding 75° abduction had no function advantage compared to the smaller fusion angles.[14]

A solid fusion is technically difficult to achieve in a child because such a large portion of the humeral head is still cartilaginous. Mah and Hall reported on a series of ten children who underwent shoulder arthrodesis for either a birth injury or poliomyelitis. All these fusions healed, and the patient reported relief of pain and satisfactory functional use of the extremity.^[5] A variety of different techniques for performing shoulder arthrodesis have been proposed using internal fixation. Vastamaki and Mah and Hall used isolated transfixing screws.[5,21] Rountree and Rockwood fixed the arthrodesis with the epiphyseal plate.[13] Rühmann et al. and Miller et al. used both plates and screws.[4,14] Makin and de Velasco Polo and Monterrubio and Charnley and Houston reported the use of Steinmann Pins, and most of them are supplemented by a spica cast. [2,11,15] These authors have reported varied results.

Rountree and Rockwood had 15 painless shoulder arthrodesis with excellent to good functional outcomes in which the abduction ranged from 90° to 45°, and they were

able to reach the head or mouth.^[13] Mah and Hall had 10 solid fusions and pain relief in 80%.[5] Miller et al., who used both plate and screws, had 10 solid fusions of 12 patients and no pain was reported, with overall satisfaction and significant improvements (abduction was average of 42.31°).[4] Rühmann et al. had 12 patients of 43 who were able to move their hands up to their faces after arthrodesis. Authors who used Steinmann pins reported 15% fusion in internal rotation.^[14]

Pooled data indicated that out of 37 patients who underwent screw arthrodesis, 16% had nonunion. Vastamaki considered it as a technical failure, due to insufficient joint debridement or inappropriate screw position.^[21] One patient had malposition with humerus fracture treated with corrective osteotomy and re-osteosynthesis. Twenty-five patients underwent a fusion with plates, 6 had nonunion, 5 infections, 6 fractures, and 2 malrotation deformities. Fifty-seven patients underwent arthrodesis with Steinmann Pins, 10.5% had pseudoarthrosis, and 5.2% had infection.

This review study on shoulder arthrodesis in pediatric and adolescent patients has multiple limitations that should be acknowledged. The heterogeneity of the patient population and the variety of surgical techniques employed make it challenging to draw definitive conclusions about the optimal approach and outcomes. In addition, the relatively small sample sizes in the included studies limit the generalizability of the findings and may introduce bias in the reported functional outcomes and complication rates. Furthermore, the variability in follow-up duration across studies may affect the assessment of long-term outcomes and complications associated with the procedure.

CONCLUSION

Arthrodesis is still a rather uncommon treatment used in a few situations to enhance shoulder function in pediatric and teenage patients when alternative reconstructive techniques would likely be ineffective. Brachial plexus damage, deltoid paralysis, rotator cuff impingement, infection, or bone deterioration are all indications of shoulder arthrodesis. Although the reported evidence on the efficacy and safety of shoulder arthrodesis is widely heterogeneous, a positive outcome was generally reported by studies regarding functional outcomes and complications.

Recommendations: In light of the complexities associated with shoulder arthrodesis in pediatric patients, it is paramount that clinicians exercise judicious decision-making regarding the timing and technique of this surgical intervention. It is recommended that arthrodesis should be performed between the ages of 5 and 9 years to maximize functional outcomes, particularly in cases of flail shoulders. The optimal positioning for shoulder arthrodesis should adhere to specific ranges, ideally between 20-30° abduction, 15-30° forward flexion, and 15-40° internal rotation, thereby avoiding hyperabduction

and excessive flexion that may compromise results. Surgeons should consider employing contemporary fixation techniques, such as the use of hybrid methods or pelvic reconstructive plates, to enhance stability and address potential bone loss. Ultimately, individualized treatment plans, grounded in the latest empirical evidence, should guide the selection of patients and techniques, ensuring the best possible recovery and functional restoration.

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Ethical approval: Since this research is a systematic review that analyzes existing literature and does not involve direct data collection from human participants, ethical approval is not applicable.

Declaration of patient consent: Patient's consent is not required, as there are no patients in this study.

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