

Natural History of the Elbow Bony Architecture in Patients With Obstetric Brachial Plexus Injury and the Association With Flexion Contractures

Eric R. Wagner, MD, MS
Jean-David Werthel, MD
Mohammad Ansari, MD
William Shaughnessy, MD
Bassem L. Elhassan, MD

From the Department of Orthopaedic Surgery, Mayo Clinic, Rochester, MN.

Correspondence to Dr. Elhassan:
Elhassan.Bassem@mayo.edu

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Abstract

Purpose: The purposes of this study were to evaluate the radiographic anatomy of the elbow and try to determine its possible relation to elbow flexion contracture in patients with obstetric brachial plexus injury (OBPI).

Methods: All patients with a history of OBPI with elbow flexion contracture who were evaluated clinically and radiographically were included in the study. A review was performed to include serial elbow examinations and previous treatment. Radiographs of the elbow were examined for the presence of bony abnormalities as a potential cause of elbow flexion contracture or the presence of progressive arthritic changes over time.

Results: Fifty-nine patients with a history of OBPI with elbow flexion contracture were included in the study. Of them, 53 had normal bony architecture, 2 had mild radial head subluxation, and 4 had chronic anterior radial head dislocations. At a mean age at final clinical follow-up of 21 years (range, 7 to 83 years), only 7% of patients had pain localized to their elbow. There were only three patients with elbow arthritis, including two of the four with radial head dislocations.

Conclusions: In the absence of a radial head dislocation, most elbow joints do not seem to undergo abnormal anatomic bony changes in patients with OBPI and flexion contractures.

Level of Evidence: Level IV (retrospective case series)

Elbow flexion contracture is common in patients with obstetric brachial plexus injury (OBPI) who do not experience full recovery of their nerve palsy. It has been reported that the incidence is up to 89.5%.¹⁻⁵ Although some combinations of soft-tissue pathologies, involving abnormal innervation and muscular imbalance, likely contribute to these contractures, the etiology underlying them is unknown. In the shoulder, internal rotation contractures have

been associated with glenohumeral deformities, such as glenoid hypoplasia, hooking of the coracoid, or varus humeral head malalignment.⁶⁻¹¹ These bony changes lead to worsening of the contractures and further limitations in function. However, it is not known whether bony and joint changes contribute to elbow flexion contractures seen in OBPI.

A paucity of information exists within the published literature on the elbow bony anatomy, its long-term

Table 1
Demographic and Surgical Data

Patients (elbows)	59 (59)
Side (right:left)	33:26
Age/follow-up (yrs)	21 (7–83)
Sex: male:female (elbows)	24:35
Previous flexion contracture surgical release	13 patients

progressive changes, and its relationship to elbow flexion contracture in patients with OBPI. The purpose of this study was to evaluate the radiographic anatomy of the elbow and its relation to elbow flexion contracture in patients with OBPI.

Methods

After approval was obtained from the institutional review board, a review was performed, examining all patients at our institution with a diagnosis of OBPI and subsequent elbow pathology from January 1, 1923, to December 31, 2005. Patients were included if they had confirmed diagnosis of OBPI in the electronic medical record, experienced and were treated at our institution for elbow flexion contractures, and underwent radiographic evaluation of the elbow for their pathology. All patients had to have a recent radiograph because we aimed at determining whether bony abnormalities and bony changes over time were reasons for elbow flexion contractures in patients with OBPI.

Clinical and Radiographic Evaluation

Clinical and radiographic analyses were performed through a review of the electronic and paper medical record. The medical record was examined to obtain other variables, including demographics, comorbidities, birth history, past surgical interventions for elbow contractures, and upper extremity function and pain. Results from the patient’s last clinic visit

were used to assess pain and elbow function. Pain was quantified as none, mild, moderate, or severe by the patients during their preoperative and postoperative interviews and standard follow-up questionnaire. Patients were also asked whether it improved or worsened throughout their lifetime. Range of motion was expressed as flexion, extension, and total arc of motion for the elbow joint. Elbow flexion strength was graded using the modified grading system of the British Medical Research Council.¹² Radiographic evaluation by three different authors (ie, E.R.W, J.-D.W, and B.T.E) was performed to determine the presence of bony abnormalities of the elbow and the presence of progressive arthritic changes over time. The radiographs were evaluated for congruency, dysplasia, and degenerative changes in the radiocapitellar, ulnotrochlear, and posterior ulnohumeral joints. Arthritic changes in the radiocapitellar and trochlear articulations were graded as none, mild, moderate, or severe. Radiographs were assessed by all three authors to either normal parameters for the age of the patient or to the patient’s noninvolved extremity when available. Subluxation was determined if the radiocapitellar line failed to pass through the capitellum.^{13,14}

Statistical Analysis

Descriptive statistics were used to summarize the data, including percentages and counts for categoric and

ordinal data and mean values and ranges for continuous data. Pain and functional outcomes and the influence of different variables on these outcomes were compared using chi-square tests (or Fisher exact tests) for categoric variables and unpaired Student *t* tests for continuous variables. All analyses were performed using the JMP statistical software package (version 8; SAS Institute). *P* < 0.05 was considered statistically significant.

Results

Demographic and Clinical Background

Over an 82-year period, we identified 59 patients who fit our inclusion criteria (Table 1). There were 35 females (59%) and 24 males. The right (compared to left) upper extremity was injured in 33 patients (55%). Every patient used their uninjured extremity as their dominant extremity. Twenty-seven patients had what appeared to be a complete birth palsy (C5-T1) with a flail shoulder, arm, and hand at birth. However, all patients recovered varying degrees of hand, elbow, and shoulder function within their first year of life. Four patients were known to have a unilateral anterior radial head dislocation that was present before evaluation at our institution, thus likely either a congenital or a long-standing developmental dislocation associated with the flexion contractures, especially given the direction of the dislocation.

Elbow Function

At a mean age at final clinical follow-up of 21 years (range, 7 to 83 years), the mean elbow flexion contracture was 30° (range, 0 to 90), the mean elbow flexion was 124° (range, 80° to 150°) of elbow flexion, and the mean total arc of motion was 94°

(40° to 130°) (Table 2). All patients had M4 or greater elbow flexion strength at final clinical follow-up. All patients were treated with therapy and splinting for their elbow flexion contracture at some point in their lives, whereas 13 patients underwent attempt at surgical soft-tissue release. The presence of a radial head dislocation did influence elbow motion because the mean elbow motion in patients with radial head dislocations was 30° to 87° (total arc of 57°) compared with 30° to 128° (total arc of 98°) in those without radial head dislocations. No other factors influenced the total elbow motion.

Elbow Pain and Limitations

Although 20 patients (34%) reported pain in their affected upper extremities, only 4 patients (7%) localized pain to their elbow. However, when pain was present, it seemed to worsen over time because it was determined during the patients' subsequent clinical visits. All patients in the study reported limitations by their injuries with daily activities and occupation. However, most reported that they were able to adapt to the elbow flexion contractures because no patient was unable to work or go to school secondary to their OBPIs.

Radiographic Evaluation

Four-view radiographs of the elbow were performed for all patients at different intervals during their clinical evaluation. As noted previously, there were four patients who experienced chronic anterior radial head dislocations that had been present for many years before their index evaluation at our institution. At a mean age at final radiologic follow-up of 16.5 years (range, 6 to 78 years), only 3 patients (5%) had radiographic evidence of moderate or severe ulnohumeral arthritis, including two of the four patients with anterior

Table 2

Clinical Outcomes and Associated Factors

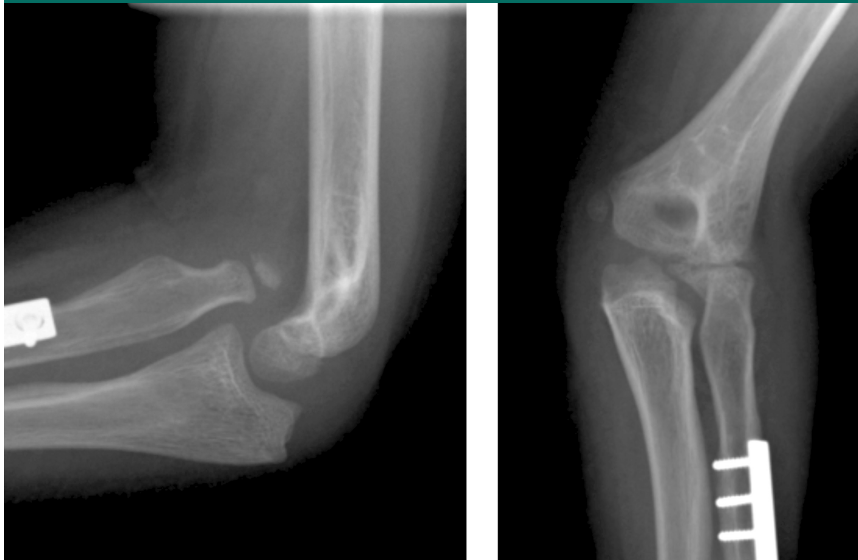
Outcome Measure	Rating
Elbow arc of motion	94° (40–130)
Elbow extension	30° (0–90)
Elbow flexion	124° (80–150)
Pain moderate/severe (patients)	
Localized to elbow	4 (7%)
Upper extremity	20 (34%)
Radiographic arthritis	
Mild	3 (5%)
Moderate/severe	3 (5%)

radial head dislocations: 25-year-old and 41-year-old women (Table 2; Figures 1 and 2). In addition to the two patients with radial head dislocations, the third patient who had advanced degenerative changes was evaluated as a 22-year-old woman with an 85° flexion contracture and no radial head dislocation, but marked limitations of her extremity from her previous OBPI. No other patients developed elbow degenerative changes. The mean flexion contracture in those with degenerative changes was 45° (95% CI, –42 to 132) compared with 29° (95% CI, 24 to 34) ($P = 0.51$). Of note, in addition to the four radial head dislocations, there were two patients who experienced mild radial head subluxation. Neither of these patients demonstrated any other abnormal radiographic findings, including normal joint and bone morphology. All four patients who had anterior radial head dislocations demonstrated proximal ulna and distal humerus dysplastic changes (ie, shallow ulnotrochlear articulation, dysplastic radial head, and abnormal radio-capitellar joint). In addition to the third patient with advanced degenerative changes without a radial head dislocation, no other elbows demonstrated any radiographic signs of dysplasia, including the two patients with mild radial head subluxation.

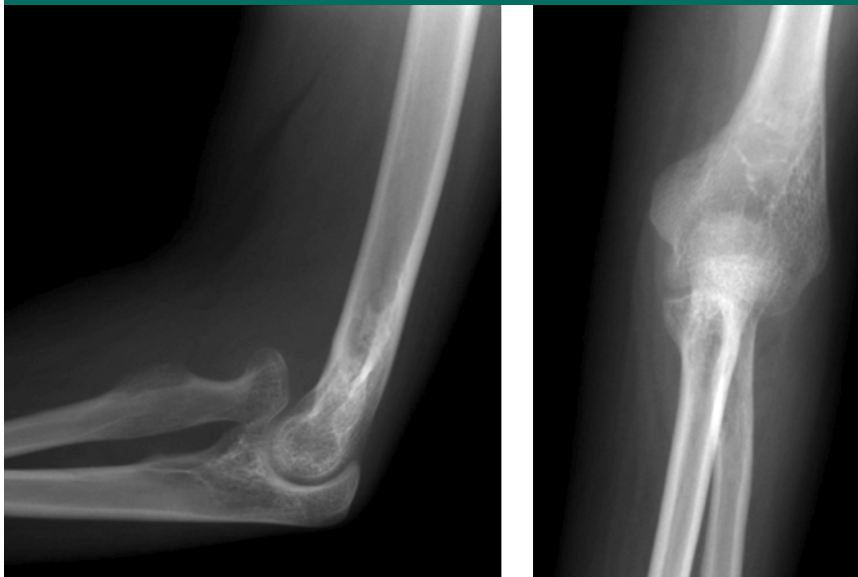
Discussion

OBPI occurs in 0.3 to 4 per 1,000 live births in developed countries.^{5,15-20} Many patients experience limitations primarily due to paralysis from muscular denervation and atrophy. These paralytic limitations are confounded by muscle contractures that progress throughout the patient's life.²¹ Elbow flexion contracture is common in patients with OBPI who did not experience early full nerve recovery. The incidence of elbow flexion contracture remains unknown, with values reported in the literature ranging from 4.6% to 89.5%.¹⁻⁵ However, the etiology underlying elbow flexion contractures remains unclear.

Although many theories exist regarding the pathogenesis of elbow flexion contractures, the exact cause leading to this common complication is still unclear. A previous report theorized that an anarchic regeneration of neurons may lead to cross-innervation of the upper limb, leading to elbow flexion contracture.⁴ In addition, Ballinger and Hoffer¹ proposed that the elbow contracture could be helpful in these patients who have weak elbow flexors to carry out many daily activities, and thus, patients were encouraged by the surgeons, families, and physiotherapists

Figure 1


Radiographs showing anterior radial head dislocation in a 10-year-old girl with a history of obstetric brachial plexus injury and an anterior radial head dislocation. Her elbow motion is limited by the flexion contracture and the radial head dislocation. Although her dislocation could be congenital, her procedure involving the radius could also have contributed to her continued dislocation.

Figure 2


Radiographs showing anterior radial head dislocation in a 38-year-old man with a history of obstetric brachial plexus injury and an anterior radial head dislocation. Over time, his radial head deformity has progressed, limiting his elbow motion and leading to signs of joint degenerative changes.

thors feel that it could simply be caused by early muscular imbalance because the elbow flexors recover earlier than the elbow extensors.^{1,3} However, this theory was contradicted by Sheffler et al,⁴ who recently showed that no correlation existed between the prevalence of elbow flexion contracture and triceps muscle weakness, suggesting that muscle imbalance might not be the cause. Finally, another hypothesis that can be found in the literature is the potential alteration in muscle contractile properties due to muscle denervation at birth.²² A recent study expanded on this theory, in which Weekley et al²³ demonstrated that denervation of the elbow flexors induced functional shortening of the elbow flexors, causing flexion contractures of variable degrees depending on the timing and extent of denervation. This same study also demonstrated that neonatal triceps tenotomy did not lead to elbow flexion contractures in their mice model. Regardless of the etiology, it seems that the soft tissues do play a critical role in the development and progression of the elbow flexion contractures in patients with OBPI.

Muscle imbalance that is known to occur in patients with OBPI leads to dysfunction of the shoulder, elbow, forearm, and wrist, mostly secondary to variable muscle paralysis, contracture, muscle imbalance, anarchic reinnervation, muscle cocontraction, and chronic joint subluxation leading to joint deformity.^{24,25} This is especially true in the shoulder in which chronic muscle imbalance and internal rotation contracture generate important glenohumeral deformities, such as glenoid dysplasia, hooking of the coracoid, or humeral head deformity.⁶⁻¹¹ Patients will often compensate for these deformities, which potentially may lead to worsening of the deformity. Although bony abnormality in the shoulder has been well established as

to remain in these flexed positions. They also proposed the idea that it could result from a persistent fetal flexion posture that was not compensated by active elbow extension in the neonatal period.¹ Other au-

possible association with internal rotation contracture of the shoulder, to our knowledge, the occurrence of bony changes in the elbow and whether these are associated with elbow flexion contractures have not previously been reported in the literature. Sheffler et al⁴ found a prevalence of 6% of radial head dislocations in a population of children with an elbow flexion contracture. However, many of the patients in their cohort had not had an elbow radiograph, suggesting that this rate of radial head dislocations might be underestimated. Furthermore, Senes²⁶ recommended resection of the olecranon as an additional surgical treatment option for elbow flexion contracture in OBPI because of the potential contribution of the bony anatomy.

In our study, we examined 59 patients with elbow flexion contractures who had undergone clinical and radiographic evaluation at our institution. At a mean clinical follow-up of 21 years (range, 7 to 83 years), only 7% of patients reported elbow pain, with a mean total arc of elbow motion of 89°. Only three patients (5%) had marked radiologic pathologies involving their elbows, two of those involving radial head dislocations. It is difficult to ascertain whether these were congenital radial head dislocations or simply long-standing dislocations associated with the flexion contractures and a “short” biceps. Regardless, although this incidence of radial head dislocation is higher than the incidence of congenital radial head dislocation observed in the general population (0.06% to 0.16%),^{27,28} this condition remains asymptomatic in most patients.²⁹ In the small number of patients with OBPI who have radial head dislocation associated with elbow flexion contracture, the radial head could be targeted in the treatment if it was felt to be contributing to the contracture.

In our series, only 3 patients (5%) had radiographic evidence of moderate or severe elbow arthritis, two of who had the anterior radial head dislocations. We tried in our study to evaluate the association between the elbow joint and flexion contractures and to determine whether any bony pathology existed that might potentially contribute to or result from the elbow flexion contractures. Our findings suggest that bony anatomic changes rarely occur in patients with OBPI, outside of those with radial head dislocations, and in this case, bony deformities are not responsible for elbow contracture.

Our study had several limitations. It is retrospective and has the inherent weaknesses of such an analysis and it is a monocentric study, which could induce a referral bias. Furthermore, the focus of this study was to examine the association of elbow bony pathology with flexion contractures. Thus, our conclusions are limited to the bony architecture, without an ability to examine the soft-tissue contributions. In addition, we recognize that using the radiocapitellar line may not predict dislocation in all pediatric cases, and occasionally, this may be a normal variant in certain pediatric elbows.^{13,14} Finally, the age range at final follow-up is from 7 to 83 years, creating a wide range of potential for arthritis development or functional abnormalities. We feel that the large range is necessary to examine the spectrum of elbow abnormalities from adolescence through adulthood. However, the strengths of this study involve its focus aim assessing the bony anatomic changes in a large number of patients over a long-term follow-up period.

In conclusion, most elbow joints do not seem to go through abnormal anatomic bony changes (eg, arthritis, dysplasia) in patients with OBPI and elbow flexion contractures, unless the patient develops anterior radial

head dislocations. This scenario indicates that flexion contracture is not primarily related to bony changes of the elbow, and in this case, surgical release of flexion contracture of the elbow should potentially focus on the soft tissues over bony or articular anatomy.

References

References printed in **bold type** are those published within the past 5 years.

1. Ballinger SG, Hoffer MM: Elbow flexion contracture in Erb's palsy. *J Child Neurol* 1994;9:209-210.
2. Yam A, Fullilove S, Sinisi M, Fox M: The supination deformity and associated deformities of the upper limb in severe birth lesions of the brachial plexus. *J Bone Joint Surg Br* 2009;91:511-516.
3. Sibinski M, Sherlock DA, Hems TE, Sharma H: Forearm rotational profile in obstetric brachial plexus injury. *J Shoulder Elbow Surg* 2007;16:784-787.
4. Sheffler LC, Lattanza L, Hagar Y, Bagley A, James MA: The prevalence, rate of progression, and treatment of elbow flexion contracture in children with brachial plexus birth palsy. *J Bone Joint Surg Am* 2012;94:403-409.
5. Aitken J: Deformity of the elbow joint as a sequel to Erb's obstetrical paralysis. *J Bone Joint Surg Br* 1952;34-B:352-365.
6. Ruchelsman DE, Grossman JAI, Price AE: Glenohumeral deformity in children with brachial plexus birth injuries. *Bull NYU Hosp Jt Dis* 2011;69:36-43.
7. Hoeksma AF, Ter Steeg AM, Dijkstra P, Nelissen RG, Beelen A, de Jong BA: Shoulder contracture and osseous deformity in obstetrical brachial plexus injuries. *J Bone Joint Surg Am* 2003;85-A:316-322.
8. Waters PM, Smith GR, Jaramillo D: Glenohumeral deformity secondary to brachial plexus birth palsy. *J Bone Joint Surg Am* 1998;80:668-677.
9. Sheehan FT, Brochard S, Behnam AJ, Alter KE: Three-dimensional humeral morphologic alterations and atrophy associated with obstetrical brachial plexus palsy. *J Shoulder Elbow Surg* 2014;23:708-719.
10. Babbitt DP, Cassidy RH: Obstetrical paralysis and dislocation of the shoulder in infancy. *J Bone Joint Surg Am* 1968;50:1447-1452.

11. Dunkerton MC: Posterior dislocation of the shoulder associated with obstetric brachial plexus palsy. *J Bone Joint Surg Br* 1989;71: 764-766.
12. Mendell JR, Florence J: Manual muscle testing. *Muscle Nerve* 1990;13(suppl): S16-S20.
13. Kunkel S, Cornwall R, Little K, Jain V, Mehlman C, Tamai J: Limitations of the radiocapitellar line for assessment of pediatric elbow radiographs. *J Pediatr Orthop* 2011;31: 628-632.
14. Ramirez RN, Ryan DD, Williams J, et al: A line drawn along the radial shaft misses the capitellum in 16% of radiographs of normal elbows. *J Pediatr Orthop* 2014;34: 763-767.
15. Pöyhä TH, Koivikko MP, Peltonen JI, Kirjavainen MO, Lamminen AE, Nietosvaara AY: Muscle changes in brachial plexus birth injury with elbow flexion contracture: An MRI study. *Pediatr Radiol* 2007;37: 173-179.
16. Foad SL, Mehlman CT, Ying J: The epidemiology of neonatal brachial plexus palsy in the United States. *J Bone Joint Surg Am* 2008;90:1258-1264.
17. Christoffersson M, Rydhstroem H: Shoulder dystocia and brachial plexus injury: A population-based study. *Gynecol Obstet Invest* 2002;53:42-47.
18. Hoeksma AF, Wolf H, Oei SL: Obstetrical brachial plexus injuries: Incidence, natural course and shoulder contracture. *Clin Rehabil* 2000;14:523-526.
19. Hardy AE: Birth injuries of the brachial plexus: Incidence and prognosis. *J Bone Joint Surg Br* 1981;63-B:98-101.
20. Evans-Jones G, Kay SPJ, Weindling AM, et al: Congenital brachial palsy: Incidence, causes, and outcome in the United Kingdom and Republic of Ireland. *Arch Dis Child Fetal Neonatal Ed* 2003;88:F185-F189.
21. Waters PM: Update on management of pediatric brachial plexus palsy. *J Pediatr Orthop B* 2005;14:233-244.
22. Stefanova-Uzunova M, Stamatova L, Gatev V: Dynamic properties of partially denervated muscle in children with brachial plexus birth palsy. *J Neurol Neurosurg Psychiatry* 1981;44:497-502.
23. Weekley H, Nikolaou S, Hu L, Eismann E, Wylie C, Cornwall R: The effects of denervation, reinnervation, and muscle imbalance on functional muscle length and elbow flexion contracture following neonatal brachial plexus injury. *J Orthop Res* 2012;30:1335-1342.
24. Gilbert A, Pivato G, Kheiralla T: Long-term results of primary repair of brachial plexus lesions in children. *Microsurgery* 2006;26: 334-342.
25. Duclos L, Gilbert A: Obstetrical palsy: Early treatment and secondary procedures. *Ann Acad Med Singapore* 1995;24: 841-845.
26. Senes F: Resection of olecranon for elbow extension. Presentation at the Club Narakas Meeting, Lausanne, Switzerland, May 23-25, 2013.
27. Kelly DW: Congenital dislocation of the radial head: Spectrum and natural history. *J Pediatr Orthop* 1981;1:295-298.
28. Mardam-Bey T, Ger E: Congenital radial head dislocation. *J Hand Surg Am* 1979;4: 316-320.
29. Bengard MJ, Calfee RP, Steffen JA, Goldfarb CA: Intermediate-term to long-term outcome of surgically and nonsurgically treated congenital, isolated radial head dislocation. *J Hand Surg Am* 2012;37: 2495-2501.