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Case Report

Congenital anterior shoulder dislocation in a newborn treated with closed reduction

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ARTICLE INFO

Article history:

Received 1 May 2018

Revised 4 June 2018

Accepted 7 June 2018

Available online 18 July 2018

Keywords:

Neonatal shoulder dislocation
Congenital shoulder dislocation
Neonatal shoulder deformity

ABSTRACT

Case: This rare case presents an isolated congenital shoulder dislocation in a twin delivery, without traumatic delivery. Delivered by emergent cesarean section at 33 weeks gestation, the infant presented with a lateral shoulder crease with x-rays showing anterior and inferior dislocation. Treatment included prompt reduction and stabilization, with follow-up ultrasound demonstrating a physeal injury.

Conclusions: This case report presents the only published congenital shoulder dislocation in an infant after an atraumatic twin cesarean delivery. Prompt reduction, stabilization, and ultrasound imaging to assess for physeal injury is our recommended management for this scenario.

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Introduction

Orthopedic injury to a neonate is a rare occurrence but usually presents with limited active motion or obvious deformity. Neonatal dislocations can be a result of birth trauma, congenital deformity, or infection. Some pertinent conditions that result in an orthopedic consultation include clavicular and humerus fractures, brachial plexus injuries, compartment syndrome, clubfoot, and joint dislocation [1].

Regardless of delivery method, any apparent flail limb or dislocation does not rule out a congenital abnormality. In the case of a flail upper extremity or shoulder deformity the most common etiologies are clavicular and humerus

fractures, or shoulder dislocation resulting in neonatal brachial plexus palsy due to shoulder dystocia. In the absence of birth trauma and neurologic deficits, a shoulder dislocation is likely congenital. Whitmann et al. [2] classified 3 types of congenital shoulder dislocations: (a) true congenital dislocation developing in utero; (b) traumatic birth directly causing dislocation; and (c) dislocation acquired secondary to a brachial plexus injury. A true congenital shoulder dislocation is considered to be the least common type.

In this case report, we present a congenital shoulder dislocation in the setting of an atraumatic birthing process. To our knowledge this is the first known case of an infant with an inferiorly displaced shoulder dislocation that was reduced successfully after an atraumatic twin cesarean delivery. This

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<https://doi.org/10.1016/j.radcr.2018.06.005>

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Fig. 1 – The picture of our patient shows the appearance of the dislocated shoulder with a noticeable lateral shoulder crease.

article discusses the method used to relocate the shoulder, the postoperative management, and a review of the literature. The authors have obtained the patient's informed written consent for print and electronic publication of the case report.

Case presentation

The mother was a 19-year-old G1 P0 with spontaneous dichorionic diamniotic twin gestation, complicated by intrauterine growth restriction of both twins. At 33 weeks, she presented for routine outpatient obstetric follow-up, and was found to have newly diagnosed absent end diastolic flow for twin B. She was subsequently admitted for urgent cesarean section, and underwent primary low transverse cesarean section with delivery of 2 twin boys. Twin B's delivery was notable for being wedged in the right cornua in transverse back down position; he was delivered with standard breech maneuvers, which involves sweeping the arm out of the hysterotomy incision using a digital pressure in the antecubital fossa; nothing remarkable was noted during deliver of the left arm, and no clicks, clunks, or pops were appreciated by the obstetricians performing the delivery. The mother recovered from this uneventfully. At the time of delivery, infants A and B had Apgar scores of 7 and 6 at 1 minute which improved to 9 and 9 at 5 minutes, respectively.

After delivery, the left arm of the twin B was noted to have a crease at the lateral aspect of the shoulder (Fig. 1) and the medical staff noticed decreased spontaneous shoulder movement compared to the contralateral side; x-rays were obtained several hours after delivery, and demonstrated an anterior

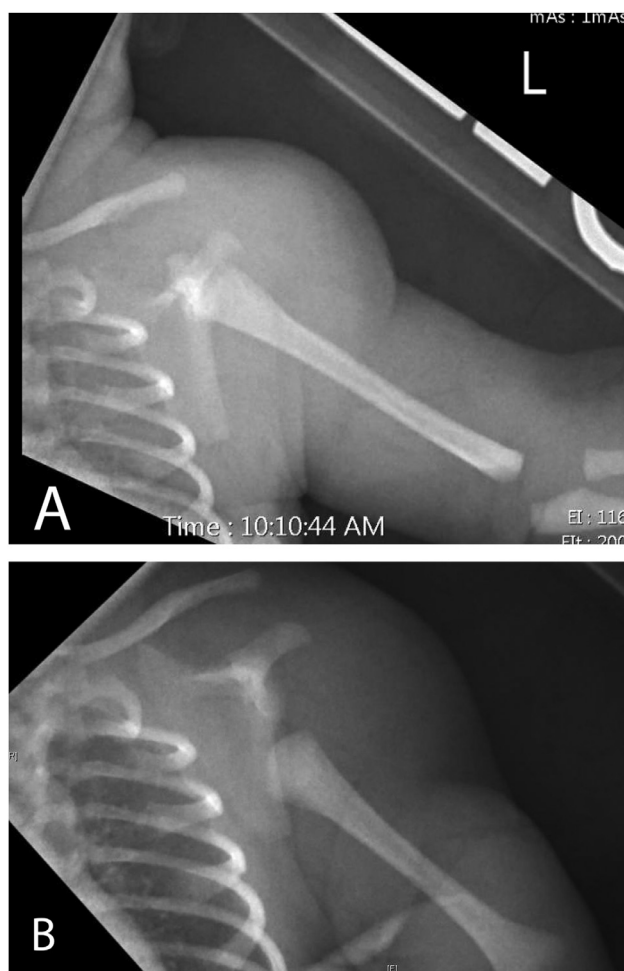


Fig. 2 – Anteroposterior (A) and lateral (B) radiograph showing a dislocated shoulder in the newborn several hours after delivery.

subcoracoid dislocation of the left shoulder with the arm externally rotated (Fig. 2A), and inferior dislocation with the arm at slight internal rotation (Fig. 2B). The contralateral shoulder was not imaged. Orthopedics was consulted to facilitate with reduction of the shoulder.

Upon initial evaluation in the Pediatric Intensive Care Unit, the left shoulder was initially abducted $\sim 120^\circ$, and he spontaneously adducted to midline without grimacing. Grip reflex was intact to the affected extremity, and the hand was pink and perfused. Gentle passive internal and external rotation was relatively well tolerated. Throughout this, there was a palpable void in the glenoid fossa, with a lateral crease just distal to the axilla, which was not present on the contralateral arm.

The reduction was initially attempted with the Kocher maneuver, which consists of elbow flexion to 90° , adduction, external rotation, and forward flexion of the shoulder to lever the humeral head into the glenoid fossa [3]. This was unsuccessful after 2 attempts, so then gentle longitudinal traction was applied via the forearm, and the shoulder was adducted with a finger in the axilla to facilitate reduction. A palpable clunk was felt, and imaging was obtained which confirmed

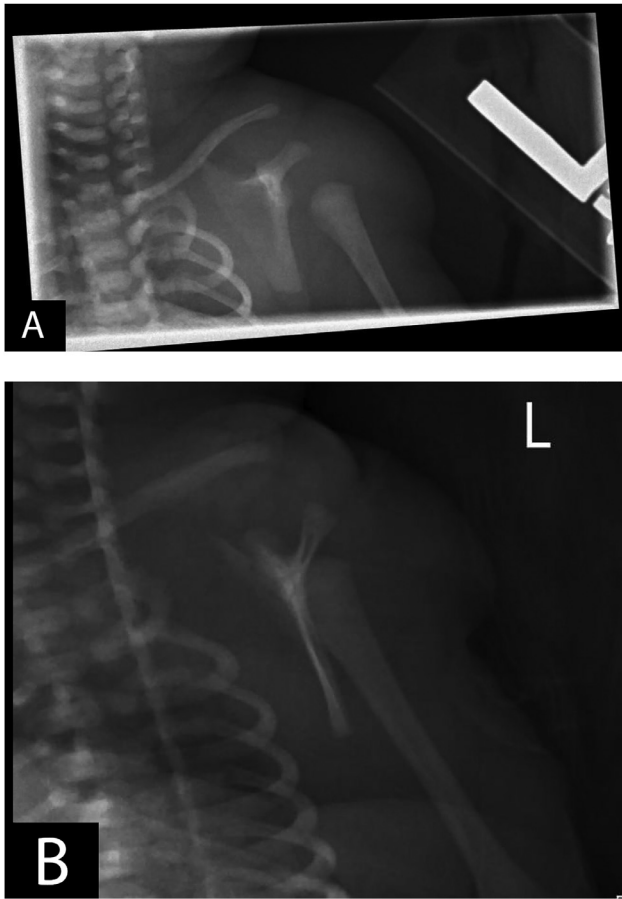


Fig. 3 – Postreduction anteroposterior (A) and lateral (B) shoulder radiographs of the newborn infant.

reduction of the shoulder (Fig. 3). Passive range of motion of the shoulder to flexion of 90°, abduction to 90°, and internal and/or external rotation arc of 90° did not result in repeat dislocation. He continued to spontaneously move the arm and maintained perfusion to the hand after this maneuver. Biceps, triceps, hand flexor and/or extensor function remained intact throughout the hospital stay.

The arm was then kept swathed in an adducted, internally rotated position for 2 weeks. Ultrasound imaging was obtained 3 days later, which demonstrated a physeal injury with slight posterior slippage of the epiphysis (Fig. 4). The ultrasound had limited visualization of the glenoid, as only the posterior portion of the glenoid was captured on a portion of the examination; there was no appreciable dysplasia of the posterior glenoid rim. At 5 months of age, the patient presented for follow-up to the pediatric orthopedic clinic and was noted to have a persistent asymmetric shoulder crease, with full symmetric range of motion to bilateral shoulders, no evidence of instability, and intact neurologic function to bilateral upper extremities. Follow-up radiographs (Fig. 5) of the left shoulder at this time demonstrated a reduced shoulder joint; radiographs of the contralateral shoulder were not obtained.

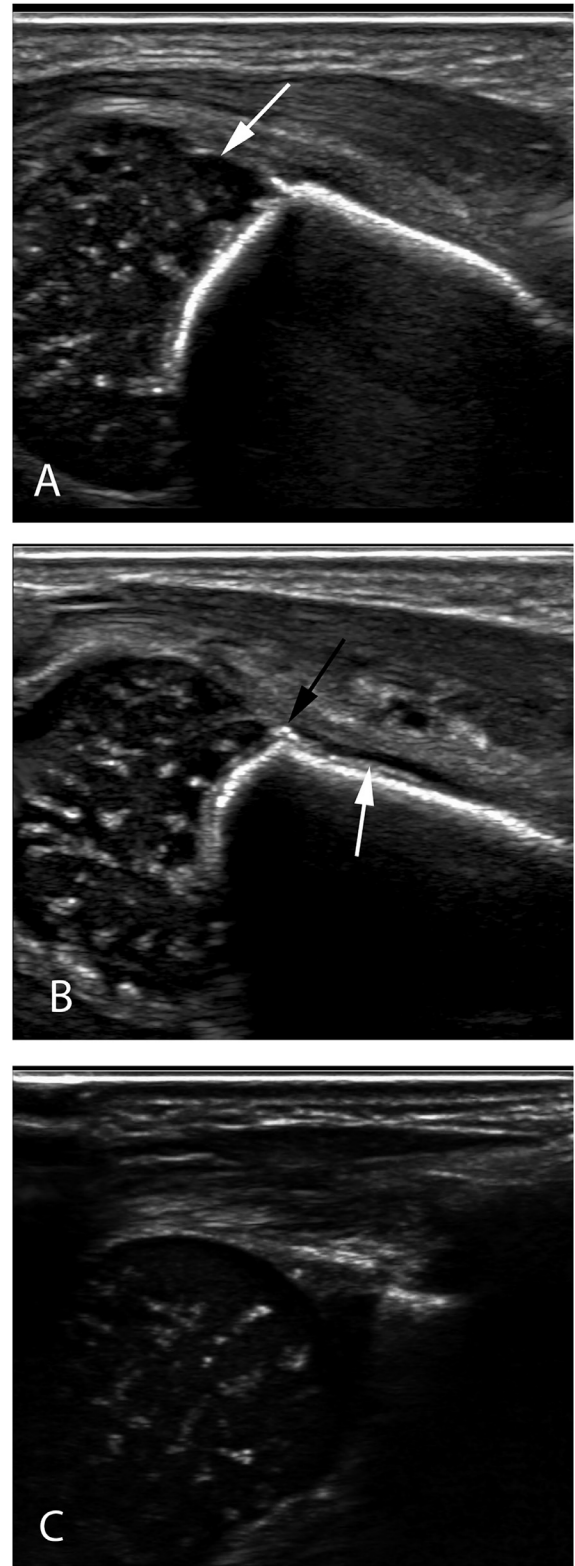


Fig. 4 – These 3 day postreduction ultrasound images show a humeral head displacement (A) with a physeal injury (B, black arrow) and periostitis (B, white arrow). (C) shows normal contour of the posterior glenoid with no appreciable dysplasia.

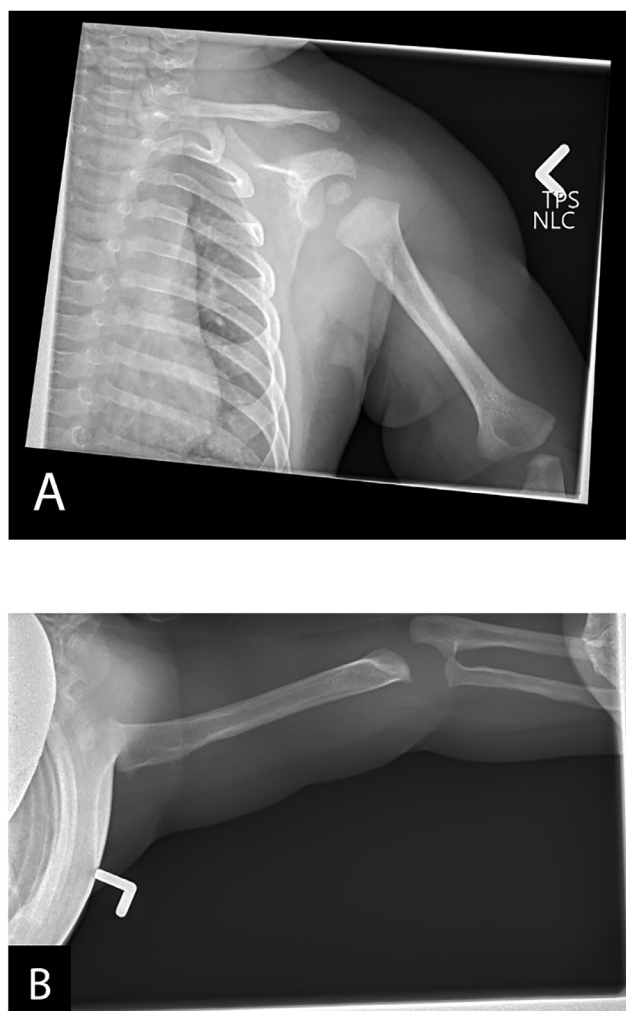


Fig. 5 – Anteroposterior and axillary shoulder x-rays at 5 months of age.

Discussion

Dislocation of the glenohumeral joint in infants is rare and usually is associated with a fracture or a neurologic problem such as a brachial plexus injury [4]. The dislocation presented in this case was not a result of obvious birth trauma, nor due to a brachial plexus injury. True congenital dislocation of the shoulder is thought to be a result of in utero maldevelopment due to bony abnormalities of the shoulder girdle [5]. Given that this shoulder was able to be reduced with a palpable clunk, and did not redislocate with provocative maneuvers, we believe that the shoulder joint developed normally and potentially dislocated shortly before birth, although another possibility is that it dislocated during delivery without being noticed. To our knowledge, this is the first known published case of a shoulder dislocation presenting at a twin birth without any reported history of birth trauma. We feel this article will serve as a useful reference for orthopedic surgeons, radiologists, and obstetricians encountering this rare pathology.

In our case there was no evidence of bony abnormalities seen on x-rays. An inferiorly dislocation in our case has only been seen in another case found in the literature, which required surgery at 6 months due to a tight contracted deltoid muscle preventing reduction [6]. Ultrasound obtained on third day of life did not demonstrate evidence of posterior glenoid dysplasia, although the ultrasound was focused more so on the proximal humerus and did not demonstrate the entire glenoid. In similar clinical scenarios to the one described above, we recommend that shoulder ultrasonography be evaluated to ensure full visualization of the proximal humerus as well as the entire glenoid, so as to rule out glenoid dysplasia as the underlying cause of dislocation.

In our literature search we found 12 true cases of congenital dislocations in infants [2,5–9]. Of these 12 patients, only 2 were cesarean deliveries [5,6]. The infant in our case had intrauterine growth restriction and was delivered prematurely by cesarean section. This minimizes the chances of a traumatic delivery. Being that this was a twin pregnancy, there could intuitively be the potential of intrauterine shoulder dislocation prior to birth due to limited space and the presence of another infant. However, a dichorionic diamniotic gestation argues against this.

Congenital dislocation of the shoulder is a very rare condition. With no history of trauma or brachial plexus injury, congenital dislocation must be considered a possible diagnosis. It is essential to make a timely diagnosis and treatment aimed at reduction and containment of the humeral head in the glenoid socket. The recommendation from this case report is that prompt reduction of the dislocation is preferable if possible by gentle manipulation. After this, we feel a short period of immobilization in a swathe is preferable. We suggest close follow-up clinically with physical examination and serial ultrasounds to confirm continued reduction of the shoulder; the added benefit of ultrasonography is the ability to identify any physeal injuries or glenoid defects. Based on the ultrasound the duration of immobilization can be modified to the specific patient.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.radcr.2018.06.005](https://doi.org/10.1016/j.radcr.2018.06.005).

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