Developmental Anterior Dislocation of the Radial Head Resulting from a Congenital Solitary Osteochondroma of the Proximal Ulna in an Infant

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Abstract
A 4-month-old female infant was brought to our office by her parents, who had noticed a lump on the child's right elbow. Examination revealed a hard, painless lump in the anterolateral region that was not reducible in flexion-extension or in pronation-supination. Neither palpation nor passive motion produced pain. Preoperative radiographs revealed a bony mass in the anterolateral region of the proximal ulnar metaphysis (solitary osteochondroma), which was displacing the radial head into anterolateral dislocation. No physiological bowing of the proximal metaphysis of the ulna was present. The infant underwent surgery at 6 months of age. No remains of the annular ligament were found. A complete resection of the tumour mass was performed, after which it was possible to reduce the radial head, together with the humeral condyle. Trans-radiocapitellar fixation was applied, with immobilization for 6 weeks. Subsequent radiological study revealed a congruent reduction of the radial head, with a progressive periosteal reaction of the posterior cortex of the ulna that evolved towards remodelling of the physiological bowing. Eight years after the surgery, the child remains asymptomatic, with complete range of motion and symmetric carrying angles. There were no relapses of osteochondroma, the deformity, or radio-ulnar synostosis.

Congenital dislocation of the radial head is a rare entity (incidence, 0.16%). Differential diagnosis of this dislocation in an infant includes several described types. It occurs either as an isolated abnormality or in association with multiple syndromes: Silver’s syndrome, Apert’s syndrome, Nievergelt’s syndrome, nail-patella syndrome, Klinefelter’s syndrome, Cornelia de Lange syndrome, and arthrogryposis, among others. The condition is usually bilateral, but some unilateral cases have been described, making it difficult to establish a differential diagnosis against a previous undiagnosed Monteggia fracture. The condition can occur both anteriorly or posteriorly, but with morphological alterations of capitellar development (absence or hypoplasia of the capitellar nucleus of ossification) or rounding of the radial head (in posterior dislocations). Echtler suggested that patients with isolated congenital dislocations of the radial head should not be treated, as 15 years later patients had only minimal functional impairment and no pain.

Abnormal bowing of the bones in the forearm can also cause a dislocation of the radial head. In rare cases, secondary radial head overgrowth and dislocation after posttraumatic dislocation of the lateral humeral condyle have been reported. These types of dislocation are developmental. Dislocations occurring after an undiagnosed Monteggia fracture, which normally are due to a green-stick fracture of the ulna, are acute. “Isolated” traumatic radial head dislocations and “isolated” congenital radial head dislocations may also occur. In case of isolated traumatic dislocation, the ulnar “bow sign,” or loss of physiological curvature of the ulna, renders reduction of the radial head impossible.

The most frequent cause of progressive dislocation of the radial head in association with osteochondroma is distal placement of the ulna, which produces alterations of the distal ulnar growth plate, causing progressive ulnar shortening and bowing of the radius, and finally to radial head dislocation. Such dislocations are usually posterior. Normal treatment of this injury involves resection of the distal osteochondroma, lengthening of the ulna, and, in some cases, hemistapling of the radial growth plate of the distal radius. All the above is associated with the general signs and symptoms of multiple osteochondromatosis.

In rare cases, the injury occurs after the development of an osteochondroma in the proximal ulna, which leads to dislocation of the radial head as a result of local compression, such as in the case described here. Dislocation of the radial head takes place in the same direction as compression (anteriorly).

Dislocation of a healthy radial head in a normal infant is exceptional. Its occurrence as a result of pressure by an osteochondroma has only been described in the literature once in children, the case of a nine-year-old boy. The presence of a congenital isolated osteochondroma makes the present case even more unique. Reasoned management of the injury is described.

Case Report

A four-month-old female infant was brought to our office by her parents, having observed a lump on the child’s right elbow. The lump had been present for at least 1.5 months. The child had no relevant previous history of illness or trauma. The parents had no relevant previous medical or surgical history, nor were there any past instances of solitary osteochondroma or multiple hereditary osteochondromata in the family. Examination revealed a hard, painless lump in the anteroexternal region of the right elbow, which was not reducible in flexion-extension or in pronation-supination. Neither palpation nor passive motion produced pain. A deficiency in passive motion was present in pronation. Active motion was insufficient for adequate evaluation, given the infant’s tender age. The preoperative radiograph revealed a bony mass in the anteroexternal region of the right elbow, which was not reducible in flexion-extension or in pronation-supination. No physiological bowing of the proximal metaphysis of the ulna was present (Fig. 1).

The infant underwent surgery at 6 months of age, following an evaluation of passive motion under anaesthesia, which revealed ranges of -50° in pronation, -10° in supination, and -15° in flexion. An external surgical approach of the right elbow was performed, using a curved, 4-cm incision between the extensor carpi ulnaris and the anconeus muscles. In the external region of the joint, the area corresponding to the radial head was found to be fully taken up by a bony mass originating in the ulna. The radial head was being constantly forced into anterior, irreducible dislocation by this osteocartilaginous mass on the proximal metaphysis of the ulna. No remains of the annular ligament were found. A complete resection of the tumour mass was performed, after which it was possible to reduce the radial head, together with the humeral condyle. Both of these elements were morphologically normal. The tumor had a cartilaginous cap with size of 3x2x1 cm. Fat taken from the subcutaneous tissue of the incision was interposed between the radial neck and the bed of the ulnar osteochondroma. Both pronation and supination were unstable, while congruency with the humeral condyle was preserved in neutral pronation-supination. Transradiocapitellar fixation was applied in that position, using a 1.2-mm Kirschner wire. The elbow was immobilized with a plaster splint, in 90° of flexion, for 6 weeks (Fig. 2). Following, the K-wire was removed and the elbow was mobilized.

Pathologic study revealed trabecular bone tissue with a highly hematopoietic marrow, covered by a layer of orderly hyaline cartilage; there were no signs of cytological abnormality. No manipulation or physiotherapy was applied to promote mobility of the elbow. Play activity with the child was the only method used to stimulate active mobility. Subsequent radiological study revealed a congruent reduction of the radial head, with a progressive periosteal reaction of the posterior cortex of the ulna (Fig. 3) that evolved towards remodelling of its physiological bowing (Fig. 4).

Six months after surgery, complete mobility was present, except for -10° in supination, which returned to normal after 9 months. Three years later, the elbow was stable and had full mobility. No relapse of the osteochondroma, the deformity,
or radioulnar synostosis had occurred. Ulnar remodelling did occur, leading to recovery of physiological curvature (Fig. 5). Eight years after surgery, the child remained asymptomatic, with complete range of motion and symmetric carrying angles. Again, no relapse of the osteochondroma, the deformity, or radioulnar synostosis, had occurred.

**Discussion**

Only one other such case has been described, that of a male child, 9 years and 6 months of age, whose injury had been present for 10 days. He was treated by means of an ulnar osteotomy and resection of the osteochondroma. Development of the osteochondroma in that case was presumably progressive throughout the growth period, since the tumor could not be described as congenital.

In contrast, in the case we have described here, development and growth of the osteochondroma undoubtedly took place during the prenatal period, since the dislocation occurred when the infant was 2 months old at most. The occurrence of a solitary osteochondroma in the prenatal period is a rarity that supports genetic etiology of the tumor (benign in isolated cases and the result of familial neoplastic syndrome in multiple cases\(^\text{11}\)) and the “congenital” description of the radial head dislocation.

Our case mimics a late, unrecognized Monteggia fracture, where the ulnar “bow sign,” or loss of physiological curvature of the ulna, renders reduction of the radial head impossible. Following reduction of the radial head, ulnar remodelling takes place, leading to recovery of physiological curvature, which had been impeded by the previous absence of the radial head from its anatomical location. Normal development of the elbow depends on the maintenance of the...
anatomic relationship between all three components of the elbow joint (ulnohumeral, radiocapitellar, and proximal radioulnar joint). Growth of the ulna and the radius takes place at the same time, and loss of proximal contact between them, therefore, produces alterations in their morphology. In the ulna, physiological curvature is the key to stability of the radial head. When absent, stability may be recovered surgically (by means of an ulnar osteotomy) or physiologically (by anatomic repositioning of both bones), which leads to a balancing of the forces they sustain and thereby stimulates remodeling of the ulnar curvature.

We agree with De Boeck that reconstruction of the annular ligament seems unnecessary, and that unreduced radial head dislocations, therefore, may be treated by simple open reduction and fixation for 6 weeks with a transarticular pin. This is why the annular ligament was not reconstructed in our case. Similarly, repair of Denécé’s square ligament was deemed to be not only impossible but unnecessary. The ligament joins the ulna and the radius in the proximal area, and was ruptured, as is the case in divergent dislocations of the elbow. The transarticular fixation procedure (temporary radiocapitellar fixation) has the drawback of potential K-wire breakage (normally within the joint), where removal of the broken fragments can be very difficult.

Although much of the literature refers to the treatment of radial head dislocation (anterior or posterior) by means of an ulnar osteotomy (ulnar flexion or extension osteotomies for the treatment of anterior and posterior dislocation of the radial head), described by Bouyala and colleagues and Hiramaya and coworkers, the main indication of this procedure is the presence of residual deformity of the ulna or radius with a concave radial head articular surface.

The rationale is the correction of the anterior bowing (or elimination of the physiological curvature) of the proximal ulna. Abe and Kayama performed an ulnar extension osteotomy in the only published case that is similar to the present one. Proximal ulnar osteotomy is standard procedure in our department to reduce chronic dislocation of the radial head in children with residual deformity of the ulna (these cases are usually neglected Monteggia fractures). In this patient, ulnar extension osteotomy was, in theory, better indicated, given the absence of curvature in the preoperative posterior cortex of the ulna. However, it was decided not to proceed with this alternative approach in order to minimize the risk of new bone formation in the proximal metaphysis of the ulna after healing of the osteotomy thereby avoiding the risk of proximal radioulnar synostosis in an area of cancellous bone exposed by resection of the osteochondroma. Furthermore, the patient’s age led us to believe that after reduction of the radial head, the forces bearing upon the humeroradial joint would forcefully induce a physiological curvature of the radius, which was indeed what occurred.

Radioulnar synostosis and loss of pronation are the major complications of late treatment of chronic radial head dislocation. Neither of these complications occurred in our case. Therefore, we agree with Kim and associates that the choice of procedure must be individualized.

We believe that in the treatment of inveterate traumatic dislocation of the radial head both methods are correct: one of them (proximal ulnar extension osteotomy) achieves a more anatomic reduction at the time, while the other (temporary radiocapitellar fixation) produces deferred reduction by progressive remodelling of the metaphysis of the ulna.

Based on this rare case, we can state that the development of a solitary, anteroexternal osteochondroma in the proximal metaphysis of the ulna leads to dislocation of the radial head and prevents anatomic development of the physiological ulnar curvature. Temporary radiocapitellar K-wire fixation of the radial head is a good additional procedure in chronic dislocations of the radial head, secondary to proximal ulnar osteochondroma. Annular ligament reconstruction is not necessary. Our experience with this case confirmed that anatomic repositioning of the radius leads to progressive physiological remodelling of the ulna.

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Disclosure Statement
None of the authors have a financial or proprietary interest in the subject matter or materials discussed, including, but not limited to, employment, consultancies, stock ownership, honoraria, and paid expert testimony.

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