Radial head dislocation due to gigantic solitary osteochondroma of the proximal ulna: case report and literature review

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Abstract Developmental anterior dislocation of the radial head resulting from a congenital solitary osteochondroma of the proximal ulna is an extremely rare condition. We present a case of a 4-year-old girl with this condition affecting her right elbow, which was treated by a trapezoidal shortening osteotomy at the radial neck following an oblique ulnar osteotomy with angulation and elongation after a complete resection of the tumor mass. The child remained asymptomatic with symmetric carrying angles during 2.5 years of follow-up post-surgery. We discuss the nature of this condition and review the literature.

Keywords Radial head dislocation · Solitary osteochondroma of the proximal ulna

Introduction

In the pediatric age group, a radial head dislocation can be congenital, post-traumatic, or developmental. Treatment approaches differ depending on the etiology. Post-traumatic dislocations should be reduced as soon as possible. On the other hand, congenital dislocation is difficult to reduce, and the majority of patients have only minimal functional impairment [3]. Developmental dislocation usually occurs due to

misalignment between the growing radius and the ulna. Prevention of a dislocation by correcting the growth discrepancy is the preferred treatment approach in these cases.

Multiple osteochondromas of the forearm occasionally disturb the bony growth and can eventually cause a radial head dislocation, which is usually treated by resection of the tumors with bone lengthening. In this case report, we describe the treatment of a female patient with a large solitary osteochondroma located at the proximal ulna, which resulted in dislocation of the radial head. Details of the surgical strategy for this patient and a review of the relevant literature are described.

Case Report

A female patient aged 3 years and 6 months with an elbow deformity was referred to our hospital by a local doctor. At 6 months of age, she presented to the local pediatric clinic with a chief complaint of a slowly enlarging mass in her right elbow. The child had no relevant previous history of illness or trauma, and the family history was negative. Examination revealed a hard, painless lump in the anterolateral region of the right elbow, which was not reducible in flexion-extension or in pronation-supination. The flexion-extension arc was 5–125° with 40° pronation and 80° supination (Fig. 1b). Serial radiographs revealed a growing bony mass in the anterolateral region of the proximal ulnar metaphysis (Fig. 1a). The proximal end of the radius was already deformed, and the radial head was dislocated in the anterolateral direction. The displacement of the radial head was progressive, but arthrography showed that the shape of the radial head was reducible (Fig. 2). The preoperative radiograph showed that the physiological bowing of the proximal metaphysis of the ulna was absent with a 3.5 mm longer radius and 6.5 mm shorter ulna than the non-affected side (Fig. 1a). Overall, the

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Fig. 1 **a** The AP and lateral radiographs of the patient at 3 years and 6 months of age. The tumor had grown at the proximal ulna, and the deformity of the dislocated proximal radius was progressive. The ulna had lost its natural posterior curvature at 6 months of age. **b** Active movement of the elbow of the patient at 3 years and 6 months of age showing limited pronation, flexion, and cubitus varus deformity. *a* extension, *b* flexion. *c* pronation

total bone length discrepancy between the radius and ulna reached 10 mm.

The patient underwent surgery at the age of 3 years and 7 months. Kocher's approach was used. The large osteocartilaginous mass protruded anterolaterally from the proximal metaphysis of the ulna, and the patient had a complete anterior dislocation of the radial head (Fig. 3). The

Fig. 2 **a** 3DCT showing a large tumor close to the radial fossa and pushing the radial head out in the anterolateral direction. **b** Arthrography revealing the hypoplastic and deformed radial head facing the flattened humeral capitellum (arrow)

Fig. 3 Intraoperative findings. **a** Exploration through the lateral incision revealed the large tumor occupying the proximal radioulnar joint. The radial head is hardly detectable. **b** After tumor resection, the deformed dislocated radial head was observed. Reduction of the dislocated radial head was impossible at that moment. *Cap* capitellum of humerus. *Asterisk* tumor. *Thick arrow* radial head. *Thin arrow* remnant of annular ligament

remnant of the annular ligament was observed (Fig. 3b). Resection of the tumor mass was performed first, and the tumor had a $2 \times 1.5 \times 1$ -cm cartilaginous cap. To correct the radioulnar length discrepancy, a 3×6 -mm trapezoidal shortening osteotomy was performed at the radial neck and then fixed with a plate (a modular hand system 1.5). The ulna was obliquely osteotomized at the proximal one third of the bone to correct the ulnar bowing. The radial head was reduced in good position, and the ulna was then rigidly fixed with the plate (a modular hand system 2.4) in a position that did not disturb the adaptation of the radiocapitellar joint through pronation-supination motion. The remnant of the annular ligament was repaired to provide additional radial head stability. The radiocapitellar stabilization was examined, and the radial head was found to be stable within the 60° of pronation and 60° of supination arc. To prevent development of radioulnar synostosis, the base of the tumor was smoothed with a surgical bar and packed with bone wax. Subcutaneous fat tissue was interposed between the radial neck and the bed of the ulnar osteochondroma. The elbow was immobilized with 90° of flexion for 3 weeks with a cylindrical cast (the hand and wrist were not immobilized) that permitted pronation-supination. Pathologic study revealed trabecular bone tissue with highly hematopoietic marrow that was covered by a layer of orderly hyaline cartilage. There were no signs of cytological abnormalities. No manipulation or physiotherapy was used to promote mobility of the elbow other than normal child play.

Subsequent radiological study revealed a congruent reduction of the radial head with a progressive periosteal reaction of the ulna that evolved towards remodeling of its physiological bowing (Fig. 4). Remodeling of the ulnar and proximal regions of the radius was observed, which led to recovery of the physiological curvature and anatomic relationship between all three components of the elbow joint (ulnohumeral, radiocapitellar, and proximal radioulnar joint). Thirty months later (Fig. 5), the elbow was stable and had complete mobility, except for 5° loss in pronation compared to the non-affected

Fig. 4 The AP and lateral radiograph at 15 months after surgery. The bony union was achieved without any complications, and the ulna shows posterior curvature with good forearm alignment

side. No relapse of the osteochondroma, deformity, or radioulnar synostosis was observed. The child remained asymptomatic with symmetric carrying angles for the 2.5 years of follow-up post-surgery. The flexion-extension arc was 5–130° with 70° pronation and 80° supination.

Discussion

A review of the literature revealed only three similar cases to the one described here [1, 4, 6]. The youngest patient was a 4-month-old infant at the initial consultation, in which a simple tumor resection with manual reduction of the radial head was successful. The radiography showed that the shape and size of the tumor was similar to our case, but the deformity was less severe because the tumor resection had been performed prior to the development of forearm bone deformity. The tumor was detected at 6 months of age in our case, and growth of the osteochondroma undoubtedly took place during the prenatal period in the previous two cases. Multiple osteochondroma are hereditary benign bone tumors, and the EXT family of tumor suppressor genes is known to be responsible for this condition. In the majority of these patients, the tumors have already appeared at birth. Although the genetic conditions for solitary osteochondroma have not been fully elucidated, reports in infants suggest that prenatal development of osteochondroma is not a rare occurrence.

The patients in the other two case reports received surgery at ages 9 and 10 years, respectively. The development of osteochondroma in these cases had been presumably progressive throughout the growth period. In the first case, the ulnar osteotomy was successful since the forearm deformity was

Fig. 5 **a** The AP and lateral radiographs of the patient at 6 years of age showing no relapse of the osteochondroma deformity. **b** Active movement of the elbow at 6 years of age showing improved range of motion and deformity. **a** flexion, **b** extension, **c** pronation, **d** supination

mild, but in the second case, the osteochondroma eventually developed into a severe forearm deformity. Although the forearm deformity was severe in our patient at the time of presentation, we believe that the patient had nearly normal anatomy before the radial head dislocation. We decided to reduce the radial head using the technique commonly used for reduction of chronic post-traumatic radial head dislocation. In order to create physiological curvature, an ulnar osteotomy was performed, and the bone length discrepancy was adjusted by an additional radial osteotomy. Radioulnar synostosis is the major complication of late treatment of chronic radial head dislocation, especially when a radial osteotomy is performed in young patients. To avoid this complication, subcutaneous fat tissue was interposed between the radius and ulna, and early forearm motion was initiated.

The necessity of annular ligament reconstruction remains controversial [2, 5]. We believe obtaining good bony

alignment is the most important aim, and the annular ligament is only a secondary constraint for stabilizing the radial head. In our case, repairing the ligament remnants was sufficient for supporting the radial head.

Benign infantile bone tumors can induce irreversible changes by disturbing the bone and joint growth year by year. However, surgical invasion may also negatively influence skeletal growth, as precise recognition of the chondral region in infants is challenging. Therefore, the decision on timing and method of treatment is difficult. In our case, since the deformation was progressive, the early surgical intervention to reposition the dislocated radial head together with tumor resection was necessary.

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Emiko Horii declares that he has no conflict of interest.

Statement of Human and Animal Rights All procedures followed were in accordance with the ethical standards of the responsible

committee on human experimentation 140 (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008 [5].

Statement of Informed Consent Identifying information, including patients' names, initials, or hospital numbers, were not published in written descriptions, photographs, and pedigrees.

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