An anomalous bifid distal ulna: A case report

C. Jones *, H. Li, N. Ellahee
Orthopaedic Department, St Helier Hospital, Wrythe Lane, Carshalton SM5 1AA, United Kingdom

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ABSTRACT

INTRODUCTION: We report a case of an osteochondroma in the form of an anomalous bifid distal ulna following mal-union of an epiphyseal injury. There has been no previous case of this reported in the medical literature.

PRESENTATION OF CASE: A 19-year-old man presented with wrist stiffness and complete loss of pronation and supination twelve months after having undergone open reduction and internal fixation for a volarly displaced distal radius fracture. Further investigation with a CT scan showed a bifid distal ulna. As this was not present on plain radiographs one year prior, it was proposed that this was an osteochondromatous growth caused by injury to the distal ulnar epiphysis. An operation was performed to excise one of the distal ulna heads, and reconstruct the TFCC to allow improved rotational movements. At one year follow-up, the patient has made an almost full recovery without complication.

DISCUSSION: We postulate that the patient sustained an occult physseal injury resulting in an osteochondromatous lesion that grew towards the joint effectively forming a second ulna head.

CONCLUSION: This is a unique case of the development of a bifid distal ulna due to physseal injury one year prior. Such a lesion has not previously been described in the distal ulna.

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1. Introduction

Spontaneous osteochondromas are the most common tumour of the skeletal system, comprising 10% of all primary bone tumours and up to 40% of benign bone tumours. Secondary osteochondromas have been commonly reported in children surviving cancer having received radiotherapy and more rarely, reports have been made hypothesising the formation of osteochondromas secondary to epiphyseal trauma. In our search of the literature we were unable to find any reported cases where such a lesion had occurred secondary to a fracture of the distal ulna with subsequent formation of a second ulna head and causing loss of movement. We report a case of a 19 year old gentleman who suffered such a complication having undergone surgical fixation of his distal radius in the same wrist one year prior.

2. Case presentation

A 19-year-old gentleman sustained a volarly displaced distal radius fracture following a motorcycle accident where he collided with another motorcycle with a closing speed of 30 mph. He was treated with a volar buttress plate (Fig. 1). The distal radio-ulnar joint was screened during the procedure and was found to be stable. The patient made an uneventful postoperative recovery.

At 6 weeks follow up radiographs showed a healing distal radius but a deformity at the distal radio-ulnar joint (DRUJ) was also noted (Fig. 2). Clinically, there was a very prominent ulna styloid with limited supination and pronation. Flexion and extension were unaffected.

Following these findings the patient was referred to an upper limb specialist for further opinion and management. At eight weeks post the original fixation, it was decided to undertake an examination under anaesthesia (EUA) with exploration/reduction of the DRUJ and removal of the buttress plate. The EUA revealed very limited supination/pronation and the subsequent arthrogram showed leakage of contrast from the DRUJ into the radio-carpal joint. The plate was successfully removed but attempted exploration of the DRUJ from the volar approach proved impossible.

Further investigations were sought in the form of a computed tomography (CT) and magnetic resonance imaging (MRI) scans. The CT scan (Fig. 3) showed an anomaly of a bifid distal end of ulna. The more volar branch was shown to be partially subluxated, with the more dorsal branch completely dorsally dislocated. Neither the distal radial or ulnar growth plates had fused allowing further growth of both ulna branches. The MRI confirmed the CT findings and commented that the triangular fibrocartilage complex (TFCC) appeared intact and was probably attached to just the smaller dorsal component with the larger more volar branch being devoid of any obvious attachment.

It was evident from the patient’s loss of function that corrective surgery would be necessary. The decision was made to re-explore

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[†] Corresponding author at: 3 Downs View Lodge, Oak Hill Road, Surbiton KT6 6EG, United Kingdom. Tel.: +44 07717363350.
E-mail address: carliw Jones@gmail.com (C. Jones).

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the DRUJ through a dorsal approach with excision of larger volar stump and a volar osteotomy of the remaining branch to bring it back into the sigmoid fossa and restore the DRUJ congruity.

However, at the time of operation it was found that volar branch was smaller and straighter than the dorsal component and articulated better mechanically with the distal radius. The larger dorsal component blocked rotation and as its preservation would have required a volar closing wedge osteotomy with ulna plating it was decided to excise it (Fig. 4). Following this, rotation was possible without impingement. The TFCC and capsule were

Fig. 1. Radiographs post injury and post operatively.

Fig. 2. Six week follow up radiograph.

Fig. 3. 3D CT reconstruction of left wrist showing Y-shaped bifid distal ulna.

Fig. 4. Intraoperative fluoroscopy left wrist post repair.
repaired. Preoperative supination and pronation were 0 and 45° respectively, increasing to 90 and 80° post-operatively.

The patient was routinely followed up post-operatively and recovered without any complications. He underwent intensive rehabilitation and two months following surgery he had a full range of flexion and extension with supination of 70° and pronation to 80°. He continued to do well post-operatively one year post reconstructive surgery.

3. Discussion

This report details the very rare case of a 19-year-old male who, following a distal radius/ulna epiphyseal fracture, developed a distal ulna osteochondroma which, although commonly reported elsewhere in the body, has not been reported in this location before. It is equally remarkable for its atypical growth characteristics in that it formed a bifid distal ulna causing progressive functional morbidity.

Osteochondromas are the most common benign bone tumour accounting for up to 50% and are commonly noted in the first three decades of life. They represent harmartomatous proliferations of bone and cartilage and are thought to arise from growth plate cartilage that has been displaced but continues to grow like an epiphysis. They may be pedunculated or sessile and have a 1% incidence of malignant transformation especially in the sessile type. They commonly occur around the knee, proximal humerus and fibula and tend to arise from and grow away from the physis mostly due to the pull of tendons and ligaments with growth continuing until skeletal maturity. Generally patients tend to be asymptomatic and the vast majority are treated conservatively although they are sometimes excised when they cause irritation to the surrounding soft tissues or become symptomatic from recurrent trauma due to their subcutaneous nature.

There have been many theories over the years postulating the origins of osteochondromas. Most of the literature relates to radiation exposure as a developmental cause. It was hypothesized by Virchow in 1891 that osteochondromas may result from separation of a cartilage fragment from the growth plate resulting in autonomous growth which is somewhat akin to the mechanism we suspect in this case.2 In a paper from 1920 put forward by Keith, the concept that a defect in the periosteal ring allowing for unrestricted growth and development lead to the development of the lesion.3

Mintzer et al. described a case in which a displaced Salter-Harris type II fracture of the distal femur subsequently developed an osteochondroma.4 One year following a closed physeal injury a small growth was noted on the lateral aspect of the femur. Subsequent radiographs over the next 5 years showed gradual increase in the size but not to the extent documented in our case. This patient’s symptoms resolved without any intervention. Greyson-Fleg et al. also reported a post traumatic osteochondroma of the distal radius in an 18 year old man, one and a half years following an undisplaced fracture at the metaphyseal-diaphyseal junction.5 In this case the patient developed an osteochondroma adjacent to the radial styloid which due to its prominence was excised.

Symptoms related to osteochondroma are usually relieved by surgery although there are no large follow-up studies in the literature and poorly defined indications for surgery. Bottner et al. reviewed 92 symptomatic osteochondromas in 86 patients (mean age 20) with 93.4% resolution of preoperative symptoms and concluded that excision was a successful form of treatment. Major complications were rare but it was noted that there was no justification for excision of asymptomatic lesions.5

With regard to the presented case we postulate that an occult ulna physeal injury sustained at the time of the distal radius fracture resulted in an osteochondromatous lesion which uncharacteristically grew towards the distal radio-ulnar joint to, in effect, form a second ulna head. Our postulation that this was trauma related and not a spontaneous case is further reinforced by the fact that the incidence of spontaneous osteochondromas in the distal radius is around 1%, and in the distal ulna much rarer.

4. Conclusion

This is a unique case of the development of a bifid distal ulna due to physeal injury one year prior. Due to the loss of supination/pronation, reconstructive surgery was undertaken and resulted in an almost complete restoration of function. Such a lesion has not previously been described in the distal ulna.

Conflict of interest

None.

Funding

None.

Ethical approval

Written consent was obtained from the patient and is available on request.

Author contributions

Carl Jones was responsible for leading the project and the writing of the case report. Harry Li was responsible for data collection and basic structuring of the report. Mr. Neil Citron and Mr. Najab Ellahwee were both involved in the early management of the patient before Mr. Citron’s untimely death. Mr. Ellahwee continued the management thereafter.

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