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Case Report

Massive osteolysis in the proximal radius and ulna due to calcium pyrophosphate dehydrate deposition disease: A case report

Yu Toda [a], Motoki Sonohata [a, *], Kazuyoshi Uchihashi [c], Hiroko Mine [b], Toshiyuki Tsuruta [b], Masaaki Mawatari [a]

[a] Departmet of Orthopaedic Surgery, Faculty of Medicine, Saga University, 5-1-1 Nabeshima, Saga 849-8501, Japan
[b] Tsuruta Orthopaedic Clinic, 1241-6, Ushidu, Saga 849-0306, Japan
[c] Departmet of Pathology, Faculty of Medicine, Saga University, Japan

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1. Introduction

The clinical manifestations of calcium pyrophosphate dehydrate crystal deposition disease (CPPD) vary widely. Many cases are found incidentally and may be asymptomatic [1]. In symptomatic patients, the lesions may be misdiagnosed as pseudorheumatoid arthritis, pseudoosteoarthritis or pseudoneuropathic joints [2,3].

CPPD is defined as a joint disease that is characterized by the presence of calcium pyrophosphate dehydrate crystals in the intraarticular and periarticular tissue [4,5]. CPPD is divided into “diffuse or non-tumoral deposition CPPD” and “massive or tumoral CPPD”. Non-tumoral CPPD is easily diagnosed by an X-ray, however, it is often difficult to make a proper diagnosis for tumoral CPPD because tumoral CPPD is the rarest clinical form of CPPD deposition.

We herein report the case of a unique clinical form of tumoral CPPD in the elbow.

The study protocol adhered to the ethical guidelines of the 1975 Declaration of Helsinki, and the study was approved by the institutional review board of Tsuruta Orthopaedic Clinic. The patient was informed that this case study would be submitted for publication, and she provided her informed consent.

2. Case report

A 77-year-old female had been diagnosed with elbow osteoarthritis at another hospital 8 years prior to admission to our hospital. Radiographs of her elbow taken at another hospital showed a slight degree of osteoarthritis with some calcareous depositions in the elbow joint (Fig. 1). She subsequently underwent an operation on her right elbow at the previous hospital (details unknown). After the operation, the pain in her right elbow improved for a few years. However, she began to notice a limited range of motion in her right elbow six years after the operation, and gradually felt pain and swelling in her right elbow. She presented to our clinic with a chief complaint of right elbow pain and swelling. The preliminary examination at the first visit to our clinic revealed swelling and pain at the right elbow. The range of motion of the right elbow was markedly decreased. The ranges of flexion/extension of the elbow motion and pronation/supination of the forearm rotation were restricted to 90°/−40° and 10°/10°, respectively. There were no subjective or objective symptoms or disorders at any other joints. The ulnar nerve disturbance was not recognized. The patient had no family history of CPPD, and no history of CPPD in other joint. The patient had no history of kidney or thyroid gland disease, and the laboratory findings, including the alkaline phosphatase and calcium levels, were within normal limits.

Radiographs of the elbow at the first visit to our clinic showed a massive mass involving the entire proximal radius, proximal ulna, and adjacent soft tissues. However, the humerus was intact. A well-defined, punctate lesion of calcification was noted in the mass. Some free bodies were observed on the anterior side of the elbow (Fig. 2). Computed tomography (CT) showed sclerotic lesions around the coronoid process of the ulna and proximal radius. Additionally, a lucent rim was observed near the lesion of the ulna and radius (Fig. 3). Magnetic resonance imaging (MRI) confirmed the presence of a space occupied lesion around the coronoid process of the ulna and proximal radius with a low-isointensity heterogeneous signal on T1-weighted and fat-saturated T2-weighted images.

The patient underwent surgical mobilization of the right elbow and resection of the calcification under general anesthesia. We

* Corresponding author. Fax: +81 952 34 2509.
E-mail address: epc9719@yahoo.co.jp (M. Sonohata).

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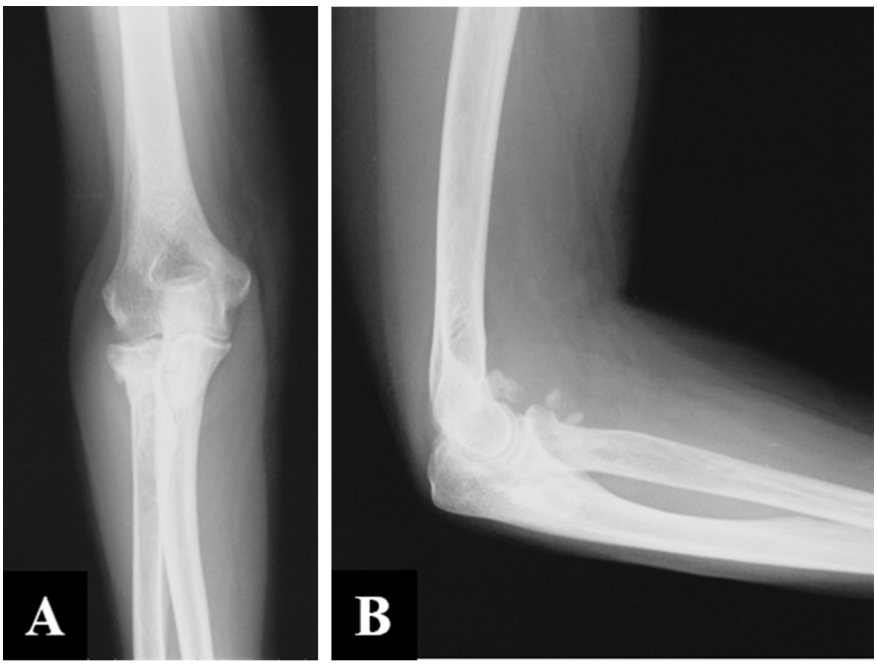

Fig. 1. Anteroposterior (**A**) and lateral (**B**) roentgenograms of the right elbow at 8 years before our operation show osteoarthritis with calcareous depositions on the anterior side of the elbow joint.

Fig. 2. Anteroposterior (**A**) and lateral (**B**) roentgenograms before our operation show a tumoral mass in the soft tissues and invading bone. A radio-opaque mass with a fluffy pattern is observed.

initially used a posterolateral approach between the anconeus and extensor carpi ulna muscles. The posterior capsule calcification was removed. The surface of the radial head and capitellum had adopted a milky white hue, due to calcium deposition. Additionally, the inside of the radial head had been filled with a chalky-white materials. As a result, we resected the radial head. The calcification on the radial side of ulna was curetted, and the cortex of radial wall of the ulna was preserved as far as possible. We then employed a medial approach. The ulna nerve was released and retracted, and the posterior oblique ligament and medial osteophytes, which had suffered from calcium deposition, were resected, while the anterior oblique ligament was preserved. Instability of the elbow was not observed after surgery. The anterior capsule was released. A milky-white mass consisting of crystals was recognized to extend from the intra capsule to the extra capsule. This mass was entirely removed. Finally, autologous bone from the ilium and artificial bone were grafted to the cavity of the proximal ulnar. Following the operation, ROM exercise was performed after 2 weeks fixation by long arm cast.

Grossly the tumor contained numerous chalky materials (Fig. 4A). Microscopically, sections showed abundant calcification and rhomboid-shaped crystals that revealed positive birefringence by polarized microscopy (Fig. 4B). The feature was compatible with CPPD.

The patient was followed up for 4 years after the operation. At the last follow-up, the range of motion of the right elbow had

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