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Forearm nonunion caused by hyperparathyroidism with 7 years follow up: A case report

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ABSTRACT

INTRODUCTION: We report a patient who developed nonunion of both bones of the forearm associated with hyperparathyroidism (HPT).
PRESENTATION OF CASE: The patient was a 71-year-old female who previously fell and hit her left hand on the ground. At 2 years after injury the patient visited our hospital, since she became aware of instability of the left forearm without an inducer due to nonunion of the radioulnar diaphysis. The patient was surgically treated to acquire forearm support. Surgery for nonunion was applied only to the ulna. To acquire an elbow joint flexion angle, an about 30° angle was added to the false joint region. At one year after surgery, blood testing suggested HPT, however, the parathyroid mass was not excised following the current guidelines for management of HPT. At 7 years after surgery, the elbow range of motion, VAS and the Q-DASH score were improved. Weight-bearing by the forearm became possible, and the patient can perform pronation and supination at the radial nonunion.
DISCUSSION: We learned from this case that it is necessary to immediately perform close examination to identify the presence or absence of primary disease causing insufficiency fracture, such as HPT. For treatment of nonunion of the 2 forearm bones in this elderly female, osteosynthesis of the ulna alone achieved sufficient osteal support without osteosynthesis of the radius, and the postoperative course was favorable.
CONCLUSION: We presented here a rare case of nonunion of both bones of the forearm associated with HPT.

1. Introduction

Hyperparathyroidism (HPT) is caused by persistent excess secretion of parathyroid hormone (PTH) by the parathyroid [1]. PTH acts to maintain the serum calcium level. In primary HPT, a mass develops in the parathyroid and causes supersecretion of PTH. Bone resorption is enhanced, and the disease may be discovered with fracture and nonunion [2]. Secondary HPT is induced by enhanced bone metabolic turnover [3], and many cases caused by renal dysfunction-associated electrolyte imbalance were reported. HPT was not discovered until nonunion or morbid fracture occurs in many cases, i.e., it is diagnosed at a stage without hypercalcemia-induced clinical symptoms [1].

On the other hand, it is very rare that nonunion of the radius and ulna simultaneously develop in the forearm. When forearm diaphyseal fracture is not appropriately treated, functional disorders of forearm support and the wrist and elbow joints may develop, and anatomical reduction serves as an important treatment index [4]. However, treatment of nonunion of the 2 forearm bones is difficult and there is no unified viewpoint. We encountered a patient in whom nonunion of the 2 forearm bones was treated with surgery for nonunion of only the ulna and achieved a favorable course. This work has been reported in line with the SCARE criteria [5].

2. Presentation of case

The patient was a 71-year-old female who fell and hit her left hand on the ground and was injured. She became aware of pain in the left forearm, visited a bone setter's office (without physician's license) and received treatment with massage. At 2 years after injury, she visited our hospital hoping for improvement of the support of the forearm. On the first examination, the range of motion of the elbow was 120° in flexion, −10° in extension, 60°

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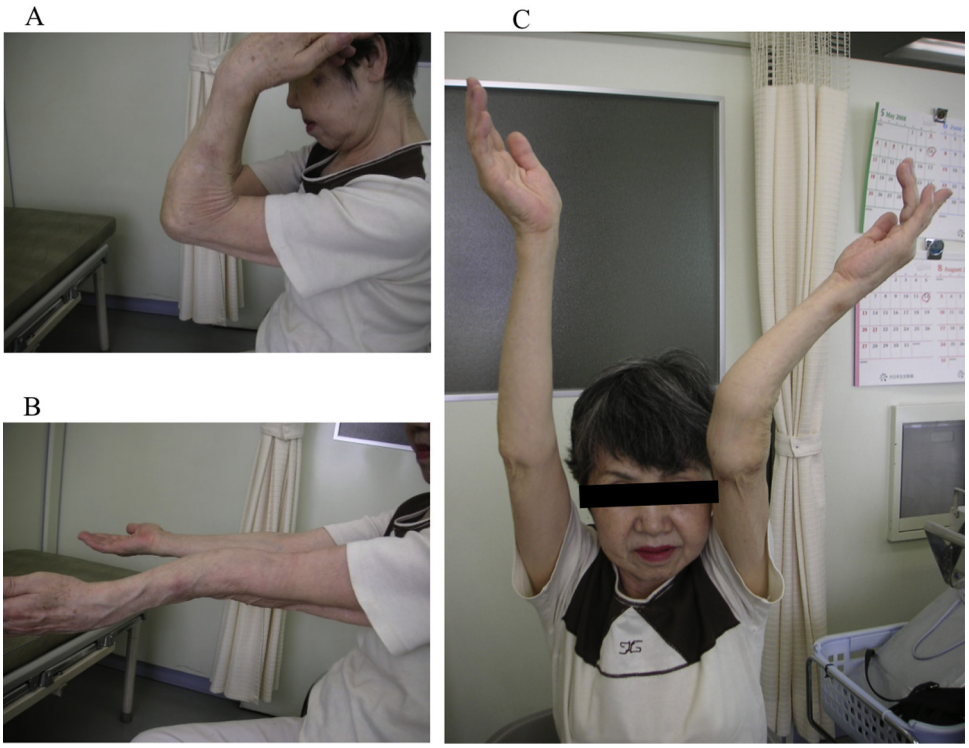

Fig. 1. Preoperative ranges of motion of the elbow. A: The range of motion of the left elbow was 100° for flexion at the first visit to our hospital. B: The range of motion of the left elbow was −10° for extension at the first visit to our hospital. C: When the patient's shoulder was elevated, she could not hold it up against her own arm weight and therefore her forearm had a curvature due to her own arm weight at the site of pseudarthrosis.

Fig. 2. Preoperative radiographs. A: The radiographic frontal view of forearm and elbow at the first visit to our hospital. B: The radiographic lateral view of forearm and elbow at the first visit to our hospital.

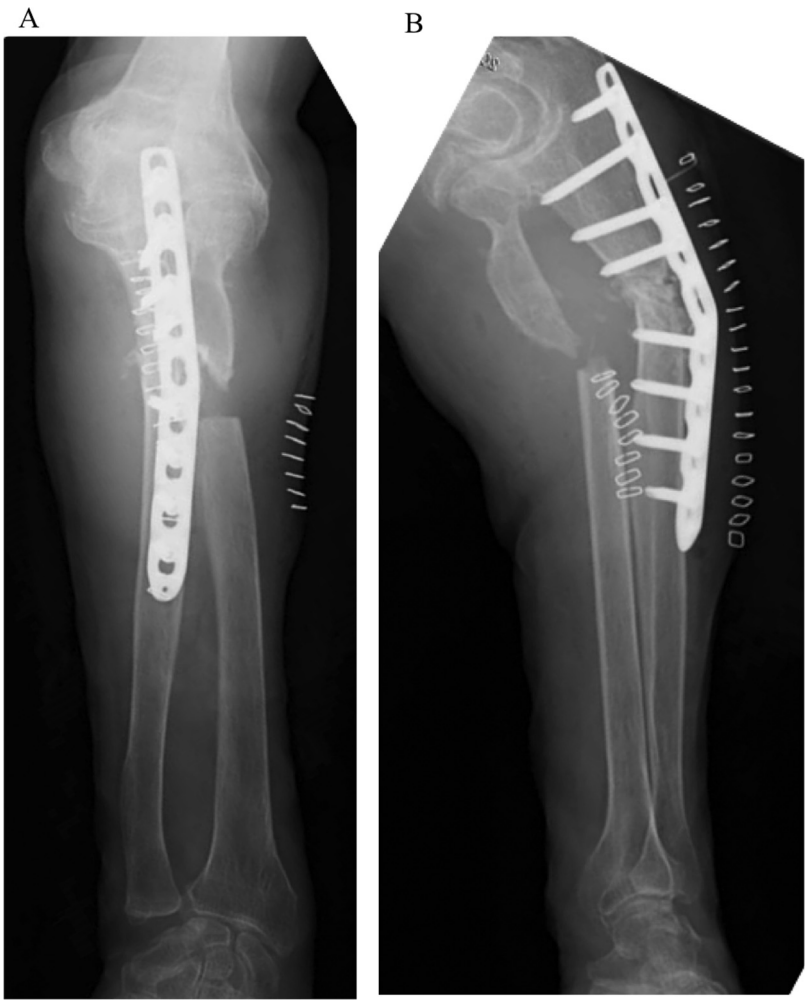

Fig. 3. Postoperative radiographs. A: The radiographic frontal view of forearm and elbow after the surgery. B: The radiographic lateral view of forearm and elbow after the surgery.

in pronation, and 60° in supination, and pronation and supination were compensated by the psedoarthrosis region (Fig. 1A and B). The psedoarthrosis region was unstable and unable to support the forearm weight, and it was about 30° curved while upper arm lifting (Fig. 1C). The grip strength was 10% of that on the health side, VAS was 5/10, and the Q-DASH score was 59.09/100. On plain radiography, the radioulnar diaphysis was nonunion, and osteoarthrosis (OA) was noted in the elbow joint (Fig. 2A and B). Based on these findings, the patient was diagnosed with functional disorder of the upper limb associated with loss of forearm support due to nonunion of the 2 forearm bones.

Surgery was performed under general anesthesia in a supine position, and the radius was reached through the dorsal Thompson approach. The distal region of the radial nonunion was shortened by resection to prevent impingement with soft tissue and the ulna during pronation and supination of the forearm. The ulna was reached through the direct posterior approach, and scar tissue of the ulnar nonunion was resected. A part of the resected radius was wedge-shaped and subjected to autologous bone grafting into the ulnar bone defect to make the ulna convex backward. A metaphyseal plate (DePuy Synthes, Tokyo, Japan) was curved by 30° backward to position the elbow joint at 135° flexion, and the ulna was fixed with this plate (Fig. 3A and B). After surgery, the forearm was immobilized with elbow joint flexion without limitation of pronation and supination for 6 weeks. Low-intensity pulsed ultrasound therapy instrument was used from 4 weeks after surgery, and bone fusion was noted 24 weeks after surgery.

On blood testing at one year after surgery, the serum Ca level was 12 mg/dL (reference range: 8.5–10.5 mg/dL), the intact PTH level was 123 pg/mL (reference range: 10–65 pg/mL), and these high levels suggested primary HPT. On ultrasonography, a mass was present in the parathyroid, based on which HPT was definitely diagnosed and bisphosphonate administration was initiated. The parathyroid mass was not excised following the current guidelines for the surgical and medical management of primary HPT [6].

At 7 years after surgery, the range of motion of the elbow was 130° in flexion, −30° in extension, 60° in pronation, and 30° in supination, VAS was 2/10, and the Q-DASH score 31.82/100, showing improvement (Fig. 4A and B). On plain radiography, bone fusion of the ulna was achieved, and the radial nonunion was maintained. Weight-bearing by the forearm became possible, and the patient can perform pronation and supination at the radial nonunion. No progression of osteoarthritis of the elbow was observed (Fig. 4C and D).

3. Discussion

The incidence of forearm nonunion is 5% or lower in many previous reports, showing that it is rare, and cases difficult to treat were also reported [7]. The most notable point which should be

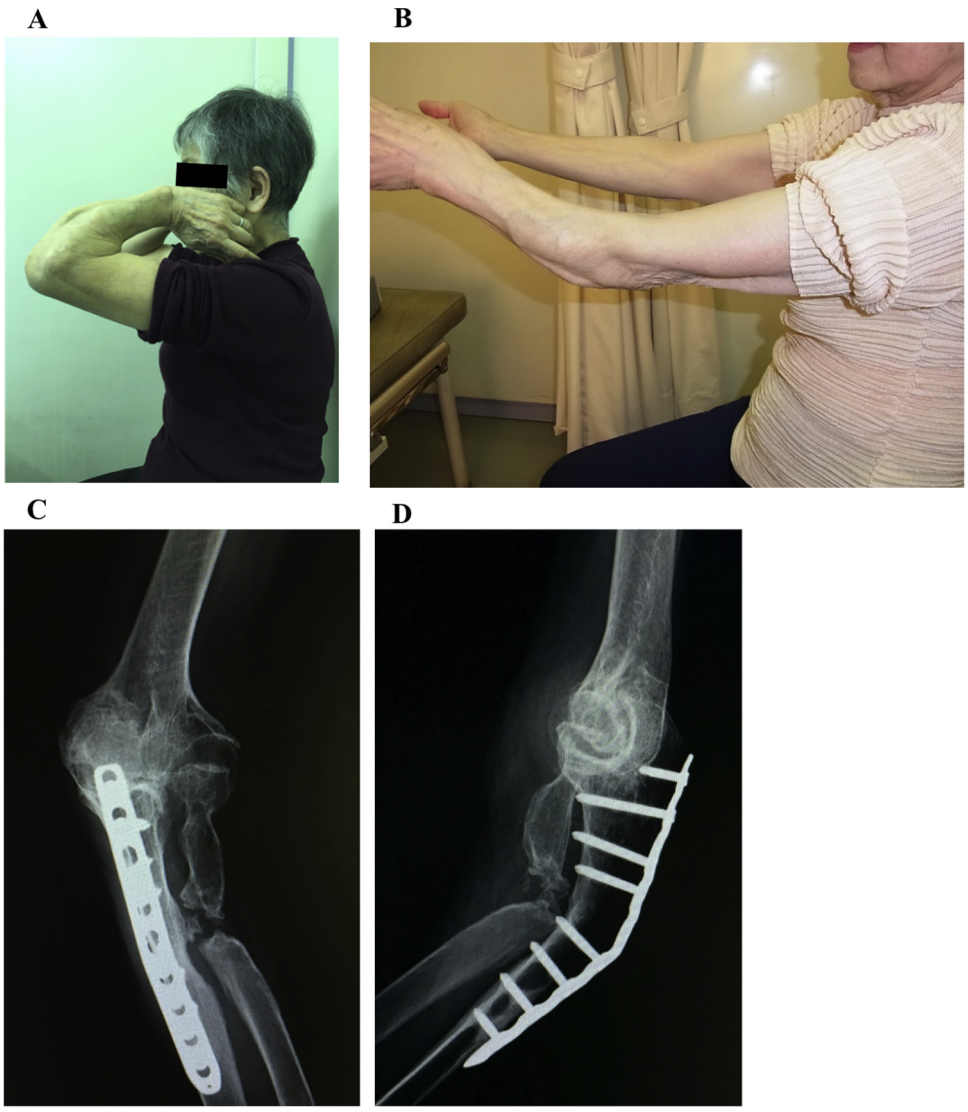

Fig. 4. Postoperative outcomes at 7 years follow-up. A: The range of motion of the left elbow was 135° for flexion at 7 years follow-up. B: The range of motion of the left elbow was −30° for extension at 7 years follow-up. C: The radiographic frontal view of forearm and elbow at 7 years follow-up. D: The radiographic lateral view of forearm and elbow at 7 years follow-up.

investigated in this patient was why nonunion developed. The development of nonunion depends on many factors, such as the fracture type, fracture region, patient background, and surgical procedure. Normally, nonunion of both forearm bones develops due to inappropriate reduction, early fixation, and inappropriate after-treatment. The present case was not appropriately diagnosed or treated because bone was fragile due to HPT and pain was mild, which may have led to the development of nonunion.

Early symptoms of HPT are nonspecific, such as listlessness, malaise, fatigue, and discomfort [8]. HPT-associated bone symptoms include osteocopic pain, reduction of BMD, insufficiency fracture of cortical bone, and in rare cases, HPT-associated bone disease [9]. Baba et al. reported that patients who developed distal radius fracture have a high risk of another insufficiency fracture, and the time suffering distal radius fracture is a good opportunity to administer therapeutic intervention to prevent the next insufficiency fracture [10]. It is no exaggeration to say that close examination of mild traumas in the elderly suspecting osteoporosis-induced insufficiency fracture is a routine work for orthopedic surgeons. Our reflection point in the present case was that osteoporosis-related close examination was performed one year after surgery. We should have closely examined the bone quality including the bone density and bone metabolism markers at the time we encountered the elderly patient with nonunion, and it may have been necessary to judge whether or not the residual bone can tolerate surgery for nonunion when nonunion was discovered. We reflected that we should have evaluated the bone quality before surgery.

Nonunion of the forearm bones is often difficult to treat [7]. In the forearm structure, the radius and ulna are lined in parallel and form the elbow joint and proximal radioulnar joint in the proximal region, and rotation of the 2 bones play an important role in these joints. When diaphyseal fracture occurs in the forearm, mobility and joint function of the forearm bones may be impaired unless it is sufficiently treated. In addition, reduction to the anatomically correct position is important, and the lengths and alignment in rotation of the 2 bones, bowing (curvature) of the radius, and the distance between the radius and ulna (interosseous space) are particularly important for maintenance of the forearm function, i.e., radioulnar fracture should be handled equivalently to intra-articular fracture because of the presence of the sophisticated motor function, anatomical function, and the positional relationship between the

radius and ulna [7]. We considered that treatment of nonunion of the 2 forearm bones aiming at bone fusion and reproduction of pronation and supination of the forearm are very complicated. Thus, in the present patient, bone fusion was applied only to the ulna, and the radius was intentionally left as nonunion. As a result, forearm support could be acquired only by the ulna, and the pronation and supination abilities of the forearm could be conserved by the false joint region of the radius similarly to the Sauvé-Kapandji procedure [11]. It was suggested that this procedure was useful for the elderly female who does not perform heavy labor. Furthermore, we performed osteosynthesis by applying a plate bent beforehand to make the ulna convex backward because her elbow joint was already with OA and flexion and extension of the elbow joint were limited. The ulna was 30° bent to make it convex backward and bone fusion was applied, expecting acquisition of 30° bending in addition to the original range of motion of the elbow.

4. Conclusion

We encountered a relatively rare case of nonunion of both forearm bones in an elderly female, and it was caused by HPT. We learned from this case that it is necessary to immediately perform close examination to identify the presence or absence of primary disease causing insufficiency fracture, such as HPT. For treatment of nonunion of the 2 forearm bones in this elderly female, osteosynthesis of the ulna alone achieved sufficient osteal support without osteosynthesis of the radius, and the postoperative course was favorable. Osteosynthesis of the ulna bend to convex backward enabled acquisition of a wider range of motion of the elbow.

Conflicts of interest

No funds were received in support of this study.

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We have no source.

Ethical approval

312, Ethical Approval of Juntendo University.

Consent

Written informed consent was obtained from patient for publication of this case report and accompanying images. A copy of the written consents are available for review by Editor-in-Chief of this journal on request.

Author contribution

All authors have contributed significantly, and that all authors are in agreement with the content of the manuscript.

Kentaro Aritomi, Kiyohito Naito, Nana NAGURA, Kenji Goto, Yoichi Sugiyama, and Yoshiyuki Iwase performed operation and ward management; Mayuko Kinoshita, Kiyohito Naito and Kazuo Kaneko diagnosed; and Mayuko Kinoshita and Kiyohito Naito wrote the paper.

Registration of research studies

researchregistry2498.

Guarantor

Dr Kiyohito NAITO.

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