Hypertrophic Pisiform: A Case of Osteoid Osteoma

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Abstract

Keywords

- ▶ osteoid osteoma
- ▶ pisiform
- ▶ hypertrophic change

We treated an extremely rare case of osteoid osteoma of the pisiform. Pisiform hypertrophy caused persistent pain and ulnar nerve irritation at Guyon's canal after the initial trauma. The re-enlargement of the pisiform attracted our attention allowing us to ultimately diagnose the condition as osteoid osteoma and treat the patient with a successful clinical result.

Osteoid osteoma is a benign bone tumor, first described by Jaffe in 1935.¹ Osteoid osteoma in the pisiform bone is especially rare, and to the best of our knowledge, only five cases have been reported before.^{2–5} One of the unique findings in this case was the hypertrophic change of the pisiform. Osteoid osteoma can cause hypertrophy of the pisiform.

Case Report

A 35-year-old male fell from a stage with his right hand outstretched. He first visited our hospital for sustained right ulnar wrist pain 9 months following the initial injury. He presented with severe tenderness of the pisiform. His pain increased with wrist ulnar deviation and extension, and was temporarily relieved with local anesthetic injection of 1% lidocaine into the pisotriquetral joint. Compression on the pisiform indicated severe pain and the triangular fibrocartilage complex stress tests was positive. Plain radiographs revealed pisiform calcification, which we diagnosed as an isolated bone island (▶ Fig. 1). Magnetic resonance imaging (MRI) indicated hydrarthrosis around the pisotriquetral joint and low signal area was found in the area of isolated bone island (▶ Fig. 2). He was treated as a case of post-traumatic pisotriquetral arthritis with oral nonsteroidal anti-inflammatory drugs (NSAIDs). Medication seemed to be effective, but the pain returned whenever he tried to discontinue NSAID medication.

As the patient's wrist pain was not well controlled, it was felt that diagnostic arthroscopy was indicated. Arthroscopy findings included a lunotriquetral tear and synovitis. However, the pisotriquetral joint could not be seen through the normal radiocarpal arthroscopy, because there is no connection between the radiocarpal and pisotriquetral joints. Arthroscopic synovectomy was performed; however, the pain gradually worsened after the operation, and started to radiate to the ulnar side of the hand, especially with the motion or touch to ulnar side of the patient's wrist. Meanwhile, he began to have swelling on the ulnar side of the wrist. Radiographs taken 10 months after the arthroscopic surgery indicated that the pisiform had enlarged (▶ Fig. 3). We could not immediately identify the reason for the enlarged pisiform, but we diagnosed Guyon's canal syndrome as the source of his pain secondary to the enlarged pisiform. The ulnar nerve was released and retracted from the enlarged pisiform at Guyon's canal, while the pisiform was trimmed 14 months after the previous surgery. The trimmed pisiform specimen was examined histologically revealing no evidence of neoplastic regions within the fragments of the bone tissue. His pain was relieved immediately after the surgery; however, in a few months, he started to complain of nocturnal pain. Radiographs taken 15 months after the second operation demonstrated the pisiform had re-enlarged, and that a radiolucent area was noted at the proximal pole of the pisiform. Computed tomography (CT) delineated a sclerotic change in the pisiform with a radiolucent

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Fig. 1 Posteroanterior (A) and oblique (B) wrist radiographs at first visit. Calcification is seen in the pisiform (arrow).

Fig. 2 Magnetic resonance imaging taken at first visit. Hydrarthrosis around the pisotriquetral joint was pointed out. Low signal area was found in the axial and coronal views at the proximal side of the pisiform. We had thought this as the isolated bone island.

Fig. 3 Radiograph taken 16 months following ▶Fig. 1. Pisiform hypertrophy is noted.

area in the proximal pole (▶Fig. 4). T1- and T2-weighted MRI indicated a low-intensity signal at the same site.

Sixteen months after the Guyon's canal release and pisiform trimming, the proximal half of the pisiform including the nidus was resected en bloc (▶Fig. 5). We chose to perform proximal partial resection, instead of the cryoablation of the nidus, to obtain the histological diagnostic confirmation. After partial resection of the hypertrophic area of the pisiform, the normal contact area of the pisotriquetral joint can be preserved not to weaken the wrist-flexion strength. Histological examination demonstrated osteoid osteoma (▶Fig. 6).

The patient experienced complete relief of symptoms immediately after the final operation. No sign of recurrence was experienced by the patient, nor was seen on radiographs 17 months after the final surgery. His wrist function was normal at the final follow-up.

Discussion

Osteoid osteoma is characterized by an osteoid-forming lesion called a nidus, reported to be less than 1.5 cm in diameter.² It accounts for ~3% of all excised primary bone tumors, and 10% of benign bone tumors.^{2,6} The patients tend to have pain, usually worse at night responding well to treatment with NSAIDs. The most common sites are the tibia and femur, and only ~2% of osteoid osteoma lesions involve the carpal bones.² The most frequently involved carpal bones are the scaphoid and the capitate, which may reflect a

Fig. 4 Computed tomography shows small radiolucent area with calcification, suggesting nidus in the sclerotic pisiform.

relationship between the size of the affected bones and the pathogenesis or ease with which to find the tumor.^{2,4} Osteoid osteoma of the pisiform is extremely rare.

Many reports have revealed a tendency for a delay in the diagnosis of osteoid osteoma of the carpal bones.¹⁻⁶ One of the reasons is that patients often present with a history of wrist injury, or other suggestive diseases such as arthritis, carpal tunnel syndrome, ganglion, infection, or neuroma. Inflammatory reaction around the nidus often causes actual wrist arthritis or synovitis. The patient may present with reduced range of motion, which is also misleading. Furthermore, the typical signs indicating presence of a nidus are difficult to see at the wrist on plain radiographs. The presence of sclerosis hides the nidus, due to the positional complexity between the carpal bones. The average time from symptom onset to the diagnosis is reported to be 18 months.²

In our case, it took almost 34 months to make the diagnosis of osteoid osteoma. The patient had a history of hand injury and actually had wrist synovitis. Also, his first two operations were both effective to some extent, and it made the diagnosis

Fig. 5 Radiographs after the third operation in which the proximal half of the pisiform including the nidus was resected.

Fig. 6 (A) Histological examination of the resected bone reveals a nidus (arrow) surrounded by sclerotic bone tissue. (B) High-power magnification of the nidus. Osteoblastic cells without cytological atypia proliferate and form irregular trabeculae of woven bone. (Hematoxylin and eosin, magnification of objective lens: a, x1; b, x10.)

even more difficult. This case illustrates how important it is to keep the rarer disease pathologies in mind, particularly when symptoms persist and/or return after treatment.

When osteoid osteoma is suspected, CT should be taken to make a correct diagnosis.² MRI has higher sensitivity in detecting a lesion; however, its contribution in showing soft-tissue changes can also lead to diagnostic error in case

of osteoid osteoma.^{2,7-10} Actually, this happened in our case too. Retrospectively evaluating this first MRI of our case, there was a T1 low and T2 low area in the body of the pisiform indicating the nidus of osteoid osteoma (►Fig. 2). Bone scan is also useful^{11,12} however, it may be difficult to differentiate the bone tumor and inflammatory changes or fracture.

Pisiform hypertrophy was characteristic in our case. A similar pisiform enlargement was noted in the first osteoid osteoma of the pisiform report.⁵ Osteoid osteoma in the cortex has been shown to cause a periosteal reaction resulting in cortex hypertrophy surrounding the nidus. This suggests that carpal bone hypertrophy can be a sign of osteoid osteoma.

In the treatment of osteoarthritis of the pisotriquetral joint, total excision of the pisiform may be recommended.¹³ However, at the third surgery of this case, we confirmed that main symptom was from the osteoid osteoma and the joint surface of the distal half of the pisotriquetral joint remained normal; thus, we excised proximal half of the pisiform to completely resection of the tumor. Successfully, patient obtained complete pain relief with normal writ function after the third operation.

Conflict of Interest

None declared.

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