Aneurysmal Bone Cyst Involving the Pisiform: A Case Report

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Abstract

Background: Aneurysmal bone cysts are an uncommon form of benign primary bone tumors; affection of the hand is very rare. **Methods:** A rigorous review of the literature showed that this type of tumor has not previously been described in the pisiform. In this article, we report the occurrence of an aneurysmal bone tumor in the pisiform of the left hand of a 19-year-old male. **Results:** This tumor was successfully treated through surgical excision with an uneventful recovery. We describe the process behind the diagnosis of the lesion and the subsequent treatment in an attempt to highlight the rare but possible occurrence of aneurysmal bone cysts in the pisiform. **Conclusion:** Appropriate treatment of aneurysmal bone cysts in this location is required, due to the anatomical nature of the pisiform itself with the risks of pathologic fracture and ulnar nerve compression.

Keywords: aneurysmal bone cyst, benign bone tumor, pisiform, hand tumors, carpal bone tumors

Introduction

First described by Jaffe and Lichtenstein,³ aneurysmal bone cysts (ABCs) are benign primary bone tumors. Although benign in nature, they can be locally aggressive, resulting in destruction of the underlying bone by the time of presentation.⁵ This lesion is uncommon, accounting for approximately 5% of primary bone tumors. Aneurysmal bone cysts mainly affect the metaphysis of long bones.^{6,8} Less than 5% of observed cases occur in the hand and usually involve the metacarpals and phalanges.² Consequently, few cases of ABCs have been reported in the carpal bones. Those affected include the capitate, hamate, triquetrum, and lunate bones.^{1,4,5,7} This article presents a case of an aneurysmal bone tumor arising in an atypical and previously unreported location, the pisiform. It demonstrates the possibility of ABC occurrence in the pisiform and the importance of considering it as a potential diagnosis when encountering painful masses in the wrist. A method of successful diagnosis and treatment of this lesion is presented.

Case Report

A 19-year-old right-handed male student presented with a mass in the left wrist. The patient complained of pain in the left wrist in daily activities. There was no pain at rest or at night. Symptoms started 6 months prior to our consult. On physical examination, a tender painful mass over the pisiform was noted. Palpation revealed a solid, bony hard, immobile in nature mass and approximately 1.8 cm × 1.5

Figure 1. Anterior posterior, lateral, and pisiform x-ray films show an expansive lytic lesion in the pisiform.

cm in size. Motor or sensory symptoms of ulnar nerve compression at Guyon's canal were negative.

Plain radiographs of the left wrist detected a radiolucent lytic lesion with bony contour expansion of the pisiform. Cortical scalloping and continuous periosteal reaction was

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Figure 2. Magnetic resonance imaging without contrast (coronal and sagittal views). Hyperintense circumscribed cystic expansive lesion involving the pisiform. No cortical destruction and soft tissue invasion were observed.

Figure 3. Intraoperative findings show a 2-cm-diameter enlarged bluish pisiform.

observed in a pisiform radiographic view (Figure 1). There was no evidence of bone collapse or pathologic fractures. Magnetic resonance imaging (MRI) showed a well-circumscribed cystic expansive pisiform lesion with high signal on T2 ruling out cortical destruction and soft tissue invasion, frequent presentation of malignant tumors in the hand. Magnetic resonance imaging report suggested a possible enchondroma (Figure 2).

Access to the tumor was achieved, using a volar approach by splitting the distal flexor carpi ulnaris (FCU). Careful dissection around the pisiform was performed protecting the neurovascular bundle at Guyon's canal at all time. Intraoperatively, an enlarged pisiform with bluish appearance was found (Figure 3). Gross and microscopic pathologic diagnosis revealed an ABC (Figure 4). Closure was performed to the FCU and subcutaneous and skin tissues. A volar splint was placed for 2 weeks until. There were no complications noted postoperatively, and patient recovery was uneventful.

Discussion

ABCs are uncommon lesions, accounting for approximately 5% of primary bone tumors. Of these, only 5% involve the bones of the hand.² Metacarpal and phalangeal involvement

Figure 4. X20 histology revealed hemorrhagic tissue with cavitary spaces divided by spindle fibroblastic cells and few giant cells. Diagnosis revealed an aneurysmal bone cyst.

is seen more commonly than carpal bone involvement. Consequently, there are only a few reported cases of ABCs to carpal bones, affecting the capitate, hamate, and lunate.^{1,4,5,7} To our knowledge, there is no literature reporting an ABC affecting the pisiform.

Although benign in nature, ABCs can behave locally aggressively, resulting in destruction of the affected bone at the time of presentation. As such, it is important to diagnose and treat these lesions promptly. Frassica et al¹ reported that intralesional curettage of the tumor and bone grafting in the hand is related to a high recurrence rate, almost 60%. Complete excision of this tumor was achieved; therefore, recurrence risk was reduced with minimal morbidity. This article aims to raise awareness of the possibility of ABC formation in the pisiform so that a similar positive outcome can be achieved in future cases.

Although rare, we believe highlighting the possibility of ABC formation in the pisiform is important and, consequently, should be included in differential diagnosis formation when encountering painful masses in the wrist. Suspicions should rise in a patient younger than 20 years old, with a primary bone tumor that presents with a lytic expansive image on the x-ray films. In the MRI, an expansive bone lesion with fluid levels can be observed without signs of malignancy or soft tissue invasion. Histology will reveal hemorrhagic tissue in cavitary spaces divided by spindle fibroblastic cells and few giant cells. The absence of malignant cells and mitotic bodies will differentiate ABCs from telangiectic osteosarcoma, a malignant bone tumor that shares age, location, and image characteristics with ABCs. The anatomical nature of the pisiform makes this location susceptible for pathologic fractures and ulnar nerve compression. Therefore, if missed or untreated, ABCs in this region could result in significant patient morbidity. Hopefully, through recognition of its pathology, this can be avoided in future cases.

Ethical Approval

As this is a case report, approval from institutional review board was not needed.

Statement of Human and Animal Rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Statement of Informed Consent

Informed consent was obtained from all individual participants included in the study.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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References

1. Frassica FJ, Amadio PC, Wold LE, Beabout JW. Aneurysmal bone cyst: clinicopathologic features and treatment of ten cases involving the hand. *J Hand Surg Am*. 1988;13:676-683.
2. Huvos AG. Aneurysmal bone cyst. In: *Bone Tumors: Diagnosis, Treatment, and Prognosis*. 2nd ed. Philadelphia, PA: W. B. Saunders; 1991;728.
3. Jaffe HL, Lichtenstein L. Solitary unicameral bone cyst, with emphasis on the roentgen picture, the pathologic appearance and the pathogenesis. *Arch Surg*. 1942;44:1004-1025.
4. Kabukcuoglu Y, Kabukcuoglu F, Kucukkaya M, Kuzgun U. Aneurysmal bone cyst in the hamate. *Am J Orthop*. 2003;32:101-102.
5. Mankin KP, Bischoff RJ, Gelberman RH, Rosenberg AE. Aneurysmal bone cyst involving the lunate. *J Hand Surg Br*. 1995;20:12-15.
6. Mirra JM, ed. *Bone Tumors: Clinical, Radiological, and Pathological Correlations*. Philadelphia, PA: Lea & Febiger; 1989:1267-1311.
7. Platt AJ, Klugman DJ. Aneurysmal bone cyst of the capitate. *J Hand Surg Br*. 1995;20:8-11.
8. Vergel De Dios AM, Bond JR, Shives TC, McLeod RA, Unni KK. Aneurysmal bone cyst: a clinicopathologic study of 238 cases. *Cancer*. 1992;69:2921-2931.