Aneurysmal Bone Cyst of the Capitate: A rare case report

Farid Najd Mazhar MD¹, Zahra Moghimi MD², Hooman Yahyazadeh MD², Sareh Shahverdi MD²

Abstract

Primary aneurysmal bone cyst (ABC) in the hands is rare. It occurs more commonly in metacarpal bones and involvement of carpal bones is very uncommon. We report the third case of ABC in the capitate, its clinical presentation, imaging findings and treatment.

Keywords: Aneurysmal bone cyst, capitate, carpal bone, hand

Cite this article as: Najd Mazhar F, Moghimi Z, Yahyazadeh H, Shahverdi S. Aneurysmal Bone Cyst of the Capitate: A rare case report. Arch Iran Med. 2014; 17(3): 211 – 214.

Introduction

A neurysmal bone cyst (ABC) known as a benign bony lesion was first introduced by Jaffe and Lichtenstein in 1942.¹ ABCs may affect any part of the skeleton, but most commonly involve the metaphysis of long bones, flat bones and vertebral column.² The occurrence of ABC in the hand are not common, accounting for nearly 3 % to 5 % of all ABCs and are often occurred in the metacarpal bones.³ ABC rarely involves carpal bones and only 2 cases of this tumor have been reported in the capitate.³ We report the third case of this tumor in the capitate along with its clinical presentation, imaging findings and treatment by curettage and bone grafting.

Case Report

A 27-year-old right handed woman was initially visited in our hand clinic with a history of 9-month pain in her right wrist. Patient was healthy otherwise. The history of trauma was negative to the right upper extremity. Pain was vague and constant and it was not exacerbated by activity.

General physical examination was normal except the findings in her right hand including mild swelling and tenderness on dorsum of the wrist. The range of motion was slightly restricted in wrist dorsiflexion.

Laboratory tests including calcium, phosphorus, alkaline phosphatase, blood count, erythrocyte sedimentation rate (ESR), and C-reactive protein (CRP) were in normal ranges.

Postero-anterior (PA) and lateral radiographs of the right wrist revealed a purely lytic lesion in the distal half of capitate (Figure 1). Whole body bone scan revealed increased uptake in the capitate area of the right wrist (Figure 2). A computed tomography (CT) scan of the right wrist showed an intraosseous cystic lesion associated with thin cortex at dorsal aspect of the capitate (Figure 3). Magnetic resonance imaging (MRI) reported a lesion

Accepted for publication: 19 November 2013

measuring 15x 8x 9 mm in the capitate, without soft tissue invasion. Signal of the lesion was high-intensity on T2-weighted and low- intensity on T1-weighted (Figure 4 a - b).

According to the imaging findings a cystic lesion like aneurysmal bone cyst was at the top of the differential diagnosis list. We approached to the lesion through a dorsal longitudinal incision. Dorsal cortex was very thin and the capitate was occupied by a blood filled cystic lesion (Figure 5). The lesion was completely evacuated by thorough curettage and specimen sent to the histologic examination (Figure 6). We filled and impacted the void area with cancellous bone graft which was harvested from the ipsilateral iliac crest.

Histopathologic examination revealed benign cystic spaces filled with blood, without endothelial lining, surrounded by fibrous septa composed of multinucleated giant cells, capillaries and spindle cell, with no nuclear atypia. The diagnosis was primary aneurysmal bone cyst (Figure 7). At 2 months of follow-up the radiographs revealed complete obliteration of the cavity with solid union and bone (Figure 8). At 12 months after surgery, the patient is asymptomatic and has no complication or recurrence, the wrist is functional, and clinical examination is normal.

Discussion

After introducing of ABC by Jaffe and Lichtenstein in 1942 for the first time, this tumor has been reported in different parts of skeleton.²

ABC is benign bone tumor with no definite etiology. opposite metaphyseal direction of blood circulation in periosteal and medullary region and trauma were proposed as the etiology of this tumor.⁴ Immuno-histochemistry and cytogenetics studies have shown that primary ABCs resemble real tumors not a reactive condition.⁵ Despite numerous investigations, the pathogenesis is still not clear and many controversies remain.⁶

ABCs predominantly affect patients under 20 years old and are more common in females and form about 1 % to 2 % of all primary bone tumors. It usually affects the metaphysis of long bones and vertebral column. In about 3 % to 5 % of cases.² ABC involves the hand rarely and it occurs most commonly in metacarpal bones.⁷

Involvement of the carpal bones by ABC is very rare and few cases have been reported in the literature.⁸⁹ To the best of our knowledge, occurrence of ABC in capitate bone have been re-

Authors' affiliations: ¹Shafa Yahyeian Rehabilitation Center, Iran University of Medical Sciences, Tehran, Iran, ²Iran University of Medical Sciences, Tehran, Iran.

[•]Corresponding author and reprints: Farid Najd Mazhar MD, ShafaYahyaian rehabilitation center, Iran University of Medical Sciences, Baharestan Sq., Tehran, Iran. Tel: +989121463048, +982133542022, Fax: +982133542020, E-Mail: fnaidmazhar@vahoo.com.



Figure 1. PA and lateral radiographs of the right wrist showing lytic lesion in the distal part of the capitate.



Figure 2. Bone scan shows increased uptake in the right wrist.

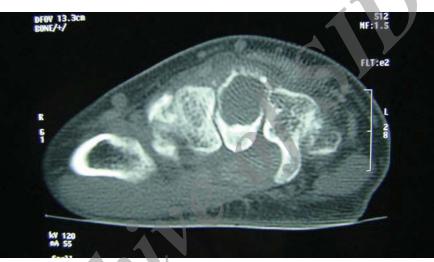


Figure 3. CT scan showing involvement of the capitate bone.



Figure 4. A) MRI T 2 weighted - showing high-intensity lesion involving the capitate bone; B) MRI T 1 weighted- low intensity lesion of the capitate bone.



Figure 5. Cystic lesion of the capitate.



Figure 6. Surgical specimen showing spongy and blood filled mass in the capitate bone.

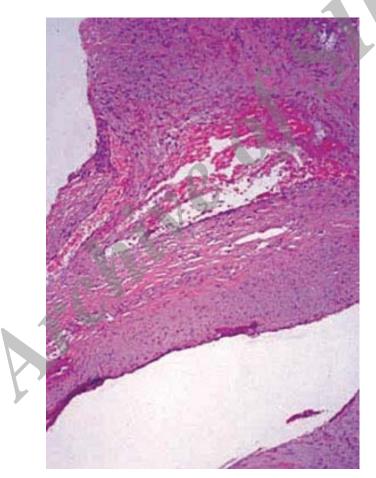


Figure 7. Histopathologic examination (H&E)- cystic spaces filled with blood, without endothelial lining, fibrous septa composed of multinucleated giant cells, capillaries and spindle cell.

ported in 2 cases previously.3,10

Carpal bone ABCs usually present with wrist pain and no specific past medical history. In most Patients imaging studies such as X-ray, CT scan and MRI reveal cystic and lytic lesion and three-phase bone scans will show increased radiotracer uptake.¹⁰ The previously reported ABC in capitate bone have occurred in 16-year-old girl and 15-year-old boy.^{3,10} Three cases(including the present case) were similar to each other in symptoms, history of pain, physical examination, imaging studies, histopathologic examination and management, but different findings were in age,



Figure 8. Follow up (2-months) radiograph of the right hand with healing the lesion.

2.

3.

4

history of trauma and area of bone involvement. In other 2 cases, patients were in common age for ABCs, they were under 20 years old, history of trauma was positive and ABCs were present at central or proximal part of capitate bone.^{3,10} In Our case the patient was a 27-year- old woman, had no history of trauma and ABC involved distal half part of her capitate bone. The wrist pain was the common presenting symptom in the mentioned cases of capitate ABC and the accepted treatment option is curettage and bone grafting which was applied in these 3 cases.

Also capitate ABC is very rare but these 3 cases indicate that this tumor should be in differential diagnosis in wrist pain especially in young patients.

Investigation was performed at: Department of Orthopedic surgery. Shafa Yahyaian Rehabilitation Center, Iran University of Medical Sciences, Tehran, Iran.

References

1. 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.

- 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.
- Sakamoto A, Tanaka K, Matsuda S, Oda Y, Tsuneyoshi M, Iwamoto Y. Aneurysmal bone cyst of the capitate: case report and a review emphasizing local recurrence. *Fukuoka Acta Med.* 2006; **97:** 302 307.
- Essadki B, Dkhissi M, Moujtahid M, Zryouil B. Diaphyseal aneurysmal bone cyst: Etio-pathogenic hypothesis and a review of the literature. *Rev Chir Orthop.* 1999; **85:** 297 – 301.
- 5. Leithner A, Machacek F, Haas OA, Lang S, Ritschl P, Radl R, Windhager R. Aneurysmal bone cyst: a hereditary disease? J Pediatr Orthop. 2004; 13(3): 214–217.
- 6. Cottalorda J, Bourelle S. Modern concepts of primary aneurysmal bone cyst. *Arch Orthop Trauma Surg*. 2007; **127**: 10 114.
- Jafari D, Jamshidi K, Najdmazhar F, Shariatzade H, Liagat O. Expansile aneurysmal bone cyst in the tubular bones of the hand treated with en bloc excision and autograft reconstruction: a report of 12 cases. J Hand Surg Eur. 2011; 36: 648 – 655.
- Lin E, Engel J, Bubis JJ, Herman O. Aneurysmal bone cyst of the hamate bone. *J Hand Surg [Am]*. 1984; 9: 847 – 850.
- 9. Mankin KP, Bischoff RJ, Gelberman RH, Rosenberg AE. . Aneurysmal bone cyst involving the lunate. *J Hand Surg [Br]*. 1995; **20:** 12–15.
- 10. Platt AJ, Klugman DJ. Aneurysmal bone cyst of the capitate. *J Hand Surg [B]*. 1995; **20**: 8 11.