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Sakamoto, Akio

Department of Orthopaedic Surgery, Graduate School of Medical Science, Kyushu University

Tanaka, Kazuhiro

Department of Orthopaedic Surgery, Graduate School of Medical Science, Kyushu University

Matsuda, Shunichi

Department of Orthopaedic Surgery, Graduate School of Medical Science, Kyushu University

Oda, Yoshinao

Department of Anatomic Pathology, Graduate School of Medical Science, Kyushu University

他

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Case Report

Aneurysmal Bone Cyst of the Capitate : A Case Report and a Review Emphasizing Local Recurrence

Akio SAKAMOTO¹⁾, Kazuhiro TANAKA¹⁾, Shuichi MATSUDA¹⁾,
Yoshinao ODA²⁾, Masazumi TSUNEYOSHI²⁾ and Yukihide IWAMOTO¹⁾

¹⁾ *Department of Orthopaedic Surgery, Graduate School of
Medical Sciences, Kyushu University, Fukuoka, Japan*

²⁾ *Department of Anatomic Pathology, Graduate School of
Medical Sciences, Kyushu University, Fukuoka, Japan*

Abstract We herein report the case of a 15-year-old boy with cystic lesion of the capitate. Magnetic resonance imaging showed that the lesion demonstrated homogenous low-intensity on T1-weighted imaging and homogenous high intensity on T2-weighted imaging. The lesion was treated by curettage and autologous bone-grafting. The histological specimen from the cystic wall showed loose fibrous tissue with capillary vessels and extravasated red blood cells. The cystic lesion was filled with blood, suggesting that the cystic lesion was actually an aneurysmal bone cyst. The present case is the second reported case of aneurysmal bone cyst of the capitate. No recurrence has been observed during the 4 years since operation. We also review the literature related to ABCs of the hand with special emphasis on local recurrence.

Key words : aneurysmal bone cyst, capitate

Introduction

Cystic lesions of the bone present a heterogeneous group of entities in which the common feature is de novo formation of unilocular or multilocular cavities filled with blood, serous fluid, mucinous contents or keratin debris⁹⁾. Cystic lesions of the carpal bones have been reported to be either aneurysmal bone cysts (ABCs) or intraosseous ganglions which are identical to its soft tissue counterparts of ganglions with the identical microscopic features. The term ABC, which is characterized by a cystic lesion filled with blood, was first

introduced in 1942¹⁴⁾. ABC has a predilection for large long bones, flat bones and vertebral bones. It has been reported that approximately 5% of ABCs are located in the bones of the hand²⁷⁾, however carpal bones are a very rare location for these lesions. In the literature, 6 cases of ABC in the carpal bones have been reported, in which there was only one report ABC arising from capitate²⁾¹¹⁾¹⁵⁾¹⁸⁾¹⁹⁾²²⁾. We encountered a case of cystic lesion in the capitate with blood as the contents, thereby suggesting that the cystic lesion was an ABC. The current case is only the second case to be reported of ABC of the capitate.

Case Report

A 15-year-old boy fell on his right hand whilst playing baseball, and a pain then

Reprint request : Akio SAKAMOTO, M.D., Ph.D.
Department of Orthopaedic Surgery, Graduate
School of Medical Sciences, Kyushu University, 3-1-1
Maidashi, Higashi-ku, Fukuoka, 812-8582, Japan
TEL +81(Japan) 92-642-5488 FAX +81(Japan) 92-
642-5507
E-mail : akio@med.kyushu-u.ac.jp

appeared in his right hand. Laboratory studies showed unremarkable findings. Radiographs showed a multilocular lesion with thinning of the cortex in the capitate. Marginal sclerosis of the lesion was evident (Fig. 1A). Magnetic resonance imaging (MRI) showed that the lesion had homogenous low-intensity on T1-weighted imaging and homogenous high-intensity on T2-weighted imaging (Figs. 1B, 1C). These homogenous signals suggested benign cystic lesion containing homogenous material. However, it was difficult further diagnostic classification. In a course of treatment, there was no tendency of healing the cystic lesion on radiographs. Therefore the lesion was treated by curettage and autologous bone-grafting. The cystic lesion was filled with blood. The his-

tological specimen from the cystic wall showed loose fibrous tissue with capillary vessels and extravasated red blood cells (Figs. 1D, 1E). The cystic wall did not have any lining cells. Reactive bone and cartilage formation were not evident. Myxoid change, which is characteristic to intraosseous ganglion, was not recognized. Any underlying condition, such as fibrous dysplasia or neoplastic lesion, was not observed. The clinicopathological features suggested a diagnosis of primary ABC. Four years after the operation, local recurrence has not been observed.

Discussion

Histologically ABCs demonstrate both septa and more solid areas which are composed of loose fibrous tissue that has numer-

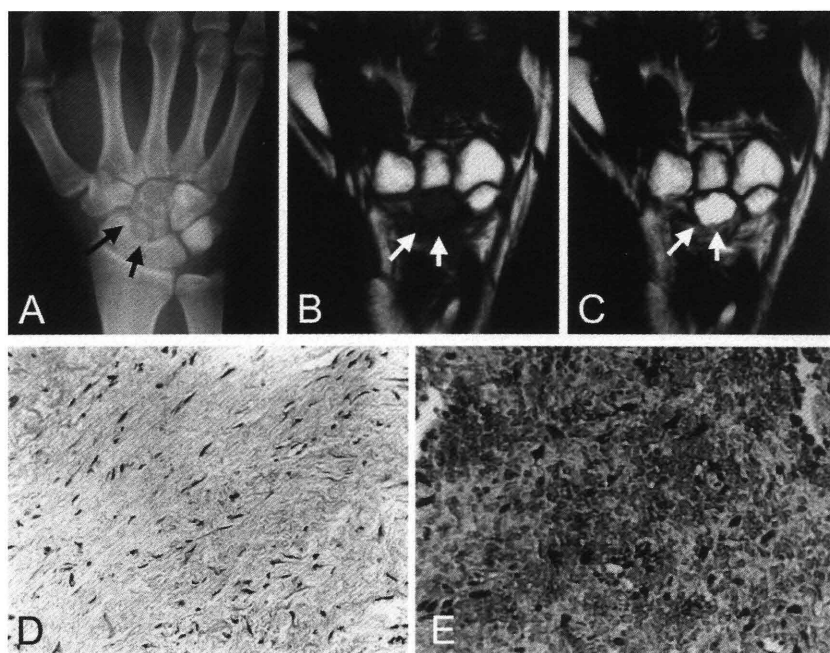


Fig. 1 Radiographs show a multilocular lesion with thinning of the cortex and marginal sclerosis in the capitate (A). MRI shows a lesion with homogenous low-intensity on T1-weighted image (B) and with homogenous high T2-weighted image (C), suggesting a benign cystic lesion. Histopathology of the cystic lesion in the capitate. The cystic wall is composed of loose fibrous tissue with capillary vessels (D). Extravasated red blood cells are prominent (E). (hematoxylin and eosin, original magnification, A : 100x, B : 300x).

ous capillary channels⁹). A solitary bone cyst (SBC) and intraosseous ganglion are necessary to be distinguished from an ABC, because they are all benign cystic bone lesions. A SBC is typically a unilocular cystic lesion that does not have a lining and which is filled with serous fluid, whereas an ABC has blood as its contents. A SBC may be confused with an ABC based on biopsy material. Similar to an ABC, a SBC is not a true neoplasm. Intraosseous ganglion is composed of a small cavity without lining that is filled with mucoid viscous material. The true incidence of this condition is unknown because many of these lesions are asymptomatic. The capsule can have two distinct layers. Characteristically, the inner layer is loose and shows myxoid change⁹). There was no such change with the specimen of the current case. The current case had blood within the cavity, and the clinicopathological features thus suggested that the current case was an ABC rather than a SBC or an intraosseous ganglion.

ABC occurs predominantly in the large long bones. In a study of 238 cases of ABC of all bones, there were 109 males (46%) and 129 females (54%), with ages ranging from 18 months to 69 years (mean, 16.1 years)²⁷). The clinical data of 41 reported ABC cases of the hand, including the present case, were available^{1~8),10~13),15~26}). They were evenly distributed with 20 males and 21 females. Affected bones were: phalanges (15 cases: 37%), metacarpal bone (19 cases: 46%) and carpal bone (7 cases: 17%) [capitate (2 cases), hamate (3 cases), trapezium (one case) and lunate (one case)]. Average age of ABCs of the hand is 20.9 years (range, 6 years to 48 years) [phalanx, 22.1 years: metacarpal bone, 18.1 years: and carpal bone, 23.0 years]. The average age of

ABCs arising in the carpal bones (23.0 years) is older than that of ABCs of all bones (16.1 years). As for the gender of the patients with ABC arising in the carpal bones, the cases were evenly distributed: 4 males and 3 females. With regard to gender, there was no significant difference between ABCs of the carpal bones and ABCs of all bones.

Over all, recurrent rates of ABCs in all bones have been reported to range from 20% to 70%⁹⁾²⁷). Recurrence in ABCs of the hand was seen in 29% of the cases (12/41). When the recurrent rate was analyzed, 53% of the phalanx cases (8/15) and 21% of the metacarpal bone cases (4/19) were recurred. On the other hand, there were no recurrent cases of the carpal bones (0/7). The small long bones such as phalange and metacarpal bones had a tendency for recurrence compared with carpal bones.

As for treatment, the most effective treatment is complete surgical excision of ABC, but in many instances such an approach may produce a major functional impairment. Therefore most lesions are treated by curettage and bone grafting⁹). Curettage with bone-grafting was undertaken in 3 cases. Excision was undertaken in 3 cases. One case of ABC in the capitate was cured after biopsy, and this case was therefore classified as having been treated by "fenestration". There were no recurrent cases in the carpal bones among these 7 cases. Although the number of reported cases is small, the prognosis of ABC in the carpal bones seems to be favorable. Interestingly, as one case was cured after biopsy alone, it is possible that fenestration could lead to a cure for this condition, but further investigation of patients with ABC in the carpal bones is necessary before making such a conclusion.

ABC is related with a blood-filled cavity

and ABC is connected with the host capillary network⁹⁾, however, the etiology of ABC is not known. ABC is likely to be initiated by a "injury" to the rich capillary network for the precursor lesion which results in an expansive destructive process caused by capillary pressure from the extravasated blood⁹⁾. ABC occurs not only as a primary condition, but also as a secondary condition associated with benign and malignant tumors. In primary, or de novo, ABCs, no underlying condition can be identified radiographically or microscopically⁹⁾. The current case had no underlying lesion, which could cause secondary ABC, suggesting that the current case was a primary ABC. It is not conclusive whether the etiology of ABC in the long bones and ABC in the carpal bones is the same, or not. The histology of the current case has less secondary changes, such as giant cell reaction, foamy cell appearance. The less secondary change might be characteristic for ABC in the carpal bones, being different from ABC in the long bones.

We herein report a case with a cystic lesion filled with blood in the capitate, suggestive of ABC. The current case is the second case of ABC in the capitate. Including the present case, a total of 7 cases of ABC of the carpal bones have been found in the literature. The clinical data of ABCs of the carpal bones such as gender were not different from those of ABCs of all bones. However, the carpal bones had a less tendency for recurrence compared with ABCs of all bones.

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References

- 1) Athanasian EA and McCormack RR: Recurrent aneurysmal bone cyst of the proximal phalanx treated with cryosurgery: a case report. *J Hand Surg [Am]* 24: 405-412, 1999.
- 2) Barbieri CH: Aneurysmal bone cyst of the hand. An unusual situation. *J Hand Surg [Br]* 9: 89-92, 1984.
- 3) Bogar WC: Aneurysmal bone cyst of the hand. *J Manipulative Physiol Ther* 16: 182-184, 1993.
- 4) Borrelli J Jr and McCormack RR Jr: Aneurysmal bone cyst involving the long finger metacarpal in a child: a case report. *J Hand Surg [Am]* 19: 800-802, 1994.
- 5) Burkhalter WE, Schroeder FC and Eversmann WW Jr: Aneurysmal bone cysts occurring in the metacarpals: a report of three cases. *J Hand Surg [Am]* 3: 579-584, 1978.
- 6) Chalmers J: Aneurysmal bone cysts of the phalanges. A report of three cases. *Hand* 13: 296-300, 1981.
- 7) Chari PR and Rao KM: Aneurysmal bone cyst of the fourth metacarpal bone. *Aust N Z J Surg* 41: 166-169, 1971.
- 8) Clough JR and Price CH: Aneurysmal bone cysts. Review of twelve cases. *J Bone Joint Surg Br* 50: 116-127, 1968.
- 9) Dorfman HD and Czerniak B: *Bone tumors*. St Louis: Mosby, 1998.
- 10) Dossing KV: Aneurysmal bone cyst of the hand. An unusual location in the first phalanges of the first finger. Case report. *Scand J Plast Reconstr Surg Hand Surg* 24: 173-175, 1990.
- 11) Frassica FJ, Amadio PC, Wold LE and Beabout JW: Aneurysmal bone cyst: clinicopathologic features and treatment of ten cases involving the hand. *J Hand Surg [Am]* 13: 676-683, 1988.
- 12) Fuhs SE and Herndon JH: Aneurysmal bone cyst involving the hand: a review and report of two cases. *J Hand Surg [Am]* 4: 152-159, 1979.
- 13) Harto-Garofalidis G, Rigopoulos C and Fragiadakis E: Aneurysmal bone cyst of the proximal phalanx of the thumb: successful replacement by tibial autograft. A case report. *Clin Orthop Relat Res* 54:

- PMID: 5589595, 1967.
- 14) Jaffe HL and Lichtenstein L: Solitary unicameral bone cyst: With emphasis on the Roentgen picture, the pathologic appearance and the pathogenesis. *Arch Surg* 44: 1004-1025, 1942.
 - 15) Kabukcuoglu Y, Kabukcuoglu F, Kucuk-kaya M and Kuzgun U: Aneurysmal bone cyst in the hamate. *Am J Orthop* 32: 101-102, 2003.
 - 16) Leeson MC, Lowry L and McCue RW: Aneurysmal bone cyst of the distal thumb phalanx. A case report and review of the literature. *Orthopedics* 11: 601-604, 1988.
 - 17) Lichtenstein L: Aneurysmal bone cyst: observations on fifty cases. *J Bone Joint Surg Am* 39A: 873-882, 1957.
 - 18) Lin E, Engel J, Bubis JJ and Herman O: Aneurysmal bone cyst of the hamate bone. *J Hand Surg [Am]* 9: 847-850, 1984.
 - 19) Mankin KP, Bischoff RJ, Gelberman RH and Rosenberg AE: Aneurysmal bone cyst involving the lunate. *J Hand Surg [Br]* 20: 12-15, 1995.
 - 20) Mortensen NH and Kuur E: Aneurysmal bone cyst of the proximal phalanx. *J Hand Surg [Br]* 15: 482-483, 1990.
 - 21) Ochsner SF and McFarland GB: Radiolucent lesion of metacarpal. *J La State Med Soc* 126: 433, 1974.
 - 22) Platt AJ and Klugman DJ: Aneurysmal bone cyst of the capitate. *J Hand Surg [Br]* 20: 8-11, 1995.
 - 23) Rao GS, Keogh P, Webster H, Lunn PG and Burke FD: Aneurysmal bone cysts in the hand treated by free non-vascular transfer of metatarsal or proximal phalanx from the foot. *J Hand Surg [Br]* 18: 736-741, 1993.
 - 24) Sakka SA and Lock M: Aneurysmal bone cyst of the terminal phalanx of the thumb in a child. *Arch Orthop Trauma Surg* 116: 119-120, 1997.
 - 25) Schwartz GB and Hammerman MZ: Aneurysmal bone cyst of the fifth metacarpal. *Orthop Rev* 18: 1309-1314, 1989.
 - 26) Slowick FA Jr, Campbell CJ and Kettelkamp DB: Aneurysmal bone cyst. An analysis of thirteen cases. *J Bone Joint Surg Am* 50: 1142-1151, 1968.
 - 27) Vergel De Dios AM, Bond JR, Shives TC, McLeod RA and Unni KK: Aneurysmal bone cyst. A clinicopathologic study of 238 cases. *Cancer* 69: 2921-2931, 1992.

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(和文抄録)

有頭骨に発生した動脈瘤性骨嚢腫の1例と文献的考察

¹⁾九州大学大学院医学研究院 整形外科学分野

²⁾九州大学大学院医学研究院 形態機能病理学分野

坂本昭夫¹⁾, 田仲和宏¹⁾, 松田秀一¹⁾,
小田義直²⁾, 恒吉正澄²⁾, 岩本幸英¹⁾

動脈瘤様骨嚢腫 (Aneurysmal bone cyst : ABC) は、血液を含む骨嚢胞性病変であり、その成因は不明である。ABC は長管骨に多くみられ手根骨発生は稀である。症例は 15 歳男性、主訴は右手関節痛。単純レントゲンにて有頭骨に辺縁骨硬化を伴う骨透亮像を認めた。MRI にて内部均一な T1 強調-低信号、T2 強調-高信号の嚢胞性病変を認めた。この病変に対して掻爬・自家骨移植術を施行した。肉眼的に病変は血液を含む嚢胞性病変であった。病理学的に

嚢胞壁は線維性組織よりなり、微小血管、血管外赤血球を認め、慢性炎症細胞出現は乏しかった。臨床病理学的所見より ABC の診断であった。術後 4 年の経過にて再発は見られていない。文献的に有頭骨発生 ABC は本症例が 2 例目であり、有頭骨を含む手根骨発生 (非長管骨) は本症例を含め 7 例の報告がある。長管骨発生 ABC の再発率は高いが、手根骨発生 ABC の再発の報告はなく、ABC の発生部位特異的な臨床所見の違いが考察された。