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Aneurysmal bone cyst of the mandible — a case report

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Aneurysmal bone cysts originating in the craniofacial region have seldom been reported in Taiwan. A case of aneurysmal bone cyst arising in the right mandible of a 15-year-old Chinese boy was presented with both the roentgenographic and microscopic findings. In view of the high recurrent rate and the large destructive mandibular lesion, segmental resection with immediate bone grafting for reconstruction and a conscientious follow up regimen was employed. The etiology and pathogenesis of this interesting lesion remained unclear but several theories were reviewed and the differential diagnosis of this lesion was also discussed. Furthermore, immunoperoxidase staining for Factor VIII related antigen was performed to help differentiate between the aneurysmal bone cyst and a central hemangioma and then to elucidate the pathogenesis of the former.

Key words: aneurysmal bone cyst.
動脈瘤性骨囊腫.

The aneurysmal bone cyst is found most commonly in long bones and in the vertebral column.[1-3] It was first recognized as a separate clinicopathologic entity by Jaffe and Lichtenstein in 1942.[4] The occurrence of this lesion in the craniofacial bones is uncommon and the first case affecting the jaw region had not been described until 1958.[5] Daugherty et al.[6] stated that aneurysmal bone cysts occurring in the craniofacial bones constituted only about 2% of all cases. A review of the English-language literature (Table 1) indicated that an aneurysmal bone cyst was a lesion occurring predominantly in young adults under 20 years of age without marked sex predilection. The oldest reported patient was a 59-year-old female while the youngest was a 4-year-old male.[8] The mandible, especially the body and ramus, was the site most frequently involved (Table 1).

To the best of our knowledge, aneurysmal bone cysts have seldom been reported in Taiwan. Owing to its lower incidence of occurrence, the aneurysmal bone cyst may be confused with other neoplastic lesions presenting with similar clinical and roentgenographic features. To avoid incorrect diagnosis, the clinicians and pathologists should be familiar with both the clinical presentations and pathologic findings of this lesion. Here in, we report an additional case of mandibular aneurysmal bone cyst with clinical, roentenographic and pathologic findings and treatment. Immunocytochemical staining for the factor VIII related antigen was also used for the differential diagnosis between the aneurysmal bone cyst and vascular tumors and for elucidating the pathogenesis of this cystic lesion.

CASE PRESENTATION

A 15-year-old Chinese male was referred to the out-patient dental clinic of Kaohsiung Medical College Teaching Hospital of the R.O.C. for the evaluation of a swelling over the right mandibular body which had lasted for 3 months. He could not be sure of a history of antecedent trauma. His past medical history and family history was non-contributory. Extraoral inspection revealed a non-tender, well-defined and bony hard swelling along the lower border of right mandibular body extending to the posterior border of the ramus of the same side (Figure 1). The overlying skin was normal in color without ulcera-

Table 1. Reported cases of aneurysmal bone cysts

Author (s)	Age	Sex	Location
Bernier & Bhaskar (1958)[5]	59	F	Mandible
Bernier & Bhaskar (1958)[5]	11	F	Mandible
Bhaskar & Bernier (1959)[7]	9	F	Mandible
Bhaskar & Bernier (1959)[7]	7	M	Mandible
Bhaskar & Bernier (1959)[7]	11	F	Mandible
Wang (1960)[30]	8	F	Maxilla
Vianna & Horizonte (1962)[3]	18	M	Maxilla
Ebling & Wangner (1964)[31]	19	M	Mandible
Lucas (1964)[32]	29	M	Mandible
Yarington, Abbott & Raines (1964)[20]	48	F	Maxilla
Koticha (1965)[33]	20	F	Mandible
Hoppe (1968)[34]	12	F	Mandible
Nosaka & Nakazana (1968)[35]	–	M	Mandible
Nobler, Higinbotham & Phillips (1968)[8]	4	M	Maxilla
Gruskin & Dahlin (1968)[26]	8	F	Mandible
Gruskin & Dahlin (1968)[26]	20	M	Maxilla
Byrd, Allen & Kindrick (1969)[36]	17	F	Maxilla
Lejeune & Burdelon (1970)[14]	17	F	Mandible
Daugherty & Eversole (1971)[6]	17	M	Maxilla
Buraczewski & Dabska (1971)[12]	26	F	Mandible
Ellis & Walters (1972)[37]	17	M	Maxilla
Komorn (1972)[26]	26	F	Maxilla
Romaniuk & Becker (1972)[39]	10	M	Maxilla
Romaniuk & Becker (1972)[39]	14	F	Mandible
Oliver (1973)[15]	20	F	Mandible
Berry, Krishan & Bhargava (1973)[40]	18	M	Mandible
Roa (1975)[41]	12	M	Maxilla
Reyneke (1978)[2]	18	M	Maxilla
Cornyn & Morris (1978)[42]	10	M	Maxilla
Hillerup & Hjorting-Hansen (1978)[43]	10	F	Mandible
Eveson, Moos, Macdonald (1978)[44]	–		Maxilla (zygoma)
Steidler, Cook & Peade (1979)[45]	21	F	Mandible
Boyd (1979)[18]	27	M	Maxilla
Boyd (1979)[18]	23	M	Maxilla
Kane (1979)[46]	14	M	Maxilla
Sastry, Wadkar & Riter (1979)[47]	–	–	Maxilla
El Deeb, Sedano & Waite (1980)[16]	19	M	Mandible
Salmo, Shukar & Albulkhail (1981)[48]	55	F	Maxilla
Saltzman (1981)[49]	22	F	Mandible
Gingell & Levy (1982)[50]	11	M	Maxilla
Gingell & Levy (1982)[50]	30	M	Mandible
Gingell & Levy (1982)[50]	14	M	Mandible
Gingell & Levy (1982)[50]	13	F	Mandible
Ueno, Mushimoto & Kurozumi (1982)[51]	13	F	Mandible
Surprenant & Tinker (1982)[52]	18	F	Mandible

Lovely (1983)[29]	14	F	Mandible
Pankey, Schaberg & Perce (1984)[53]	11	M	Mandible
Hempenstall, Campbell, Radden & Reade (1984)[54]	24	F	Mandible
Robinson (1985)[55]	13	M	Maxilla
Zachariades & Vairaktaris (1986)[56]	35	F	Maxilla
Zachariades & Vairaktaris (1986)[56]	37	M	Mandible
Eisenbud, Attie, Gaslick & Platt (1987)[25]	48	M	Mandible
Toljanic & Lechewski (1987)[57]	16	M	Mandible
Nadimi, Brony & Sbigoli (1987)[22]	38	M	Maxilla
Newman (1987)[58]	17	M	Mandible
Nik Noriah Nik-Hussein & Boon (1989)[59]	7	M	Mandible
Giddings, Kennedy & Smith (1989)[60]	14	M	Mandible
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Carmichael, Malcolm & Ord (1989)[61]	12	M	Maxilla (zygoma)

tion. One lymph node was palpable at the right submandibular area, which was tender upon palpation. The intraoral examination showed a normal oral condition and a well-cared dentition. No percussion pain was noted for teeth #29, #30 and #31 and all three teeth and their contralateral counterparts revealed normal vitality by the electric pulp test.

Roentgenographic examination showed an expansile osteolytic lesion extruding from the lower border of the right mandibular body with periosteal reaction (Figure 2). Computed tomography revealed an osteolytic lesion with soft tissue density which occupied the right mandibular body extending to the angle area homolaterally (Figure 3).

An incisional biopsy of the lesion under

Figure 1. Preoperative view showing a right submandibular swelling (arrowhead).

Figure 2. Preoperative panoramic radiograph of right mandible showing an expansile lesion extruding from the lower border of the mandible with periosteal reaction (arrowhead).

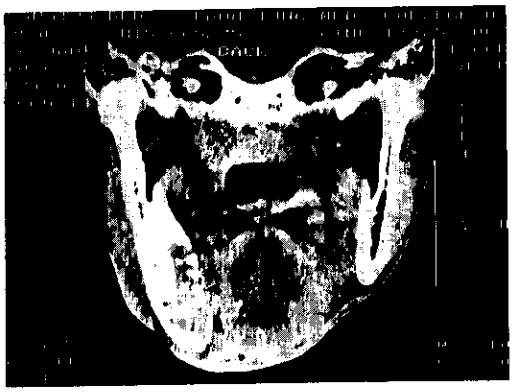

Figure 3. CT scan showing an expansile osteolytic lesion of the right mandibular body (arrowheads).

local anesthesia was performed via an extraoral approach. The right submandibular lymph node was also excised. After removing the paper-thin like expaned cortical bone, many blood filled cystic cavities separated by a thin septa were noticed. Bleeding of a pulsating type was observed and hemostasis was achieved with difficulty. The cavities was packed with a tampon and the wound was closed. The patient was then admitted to our dental ward for the observation of any uncontrollable bleeding after the incisional biopsy and was scheduled for surgery. The pathologic report of the incisional biopsy was consistent for an aneurysmal bone cyst and lymphoid hyperplasia of the right submandibular lymph node. The results of the complete blood and urine examinations were within the normal range with the exception of the alkaline phosphatase which showed an elevated value of 213 IU/1 (normal range: 26.0 - 88.0 IU/1) indicating a possible active osteoblastic and/or osteoclastic activity. Under endotracheal general anaesthesia, segmental resection from the right second mandibular premolar to the ramus of the same side was done. The bone gap resulting from the mandibular resection was bridged by a graft obtained from the right iliac crest which was fixed by stainless-steel miniplates. Following the placement of the bone graft, the submandibular wound was closed in layers. Intermaxillo-mandibular fixation was applied and the occlusion was restored. The postoperative course was uneventful.

Grossly, the resected mandibular specimen measuring about 7 cm in diameter consisted of the right portion of mandible with the associated teeth #29, #30, #31 and revealed a ballooned-out appearance with greatly thinned cortical bone resembling parchment. Upon the removal of the cortical plate, multiple spiderweb-like compartments of varied sizes filled with dark red blood coagulum were observed.

Microscopically, hematoxylin & eosin stained sections showed multiple sinusoidal blood-filled cavities within the fibrous connective tissue in which multinucleated giant cells were scattered and foci of reactive osteoid arranged in a circumscribing distribution were found (Figure 4). Furthermore, small and large venous lakes within a fibrovascular stroma could be observed (Figure 5).

Immunohistochemical staining for Factor VIII related antigen (F VIII RAg), at the concentration of 1:300, by the peroxidase-anti-peroxidase (PAP) method showed that the surface of sinusoidal blood spaces was exclusively devoid of endothelium (Figure 6). However, antibodies to F VIII RAg reacted with their appropriate antigens in the walls of blood vessels which served as internal controls

Figure 4. Microphotograph showing a typical sinusoidal space filled with blood coagulum (left bottom corner) and reactive trabeculae of bones(B) present in the fibrous stroma with scattered multinucleated giant cells (right bottom corner). (H & E, x100)

Figure 5. Microphotograph showing many small and large venous lakes (V) within a fibrovascular stroma. A sinusoidal space (S) is at the left bottom corner. (H & E, x100)

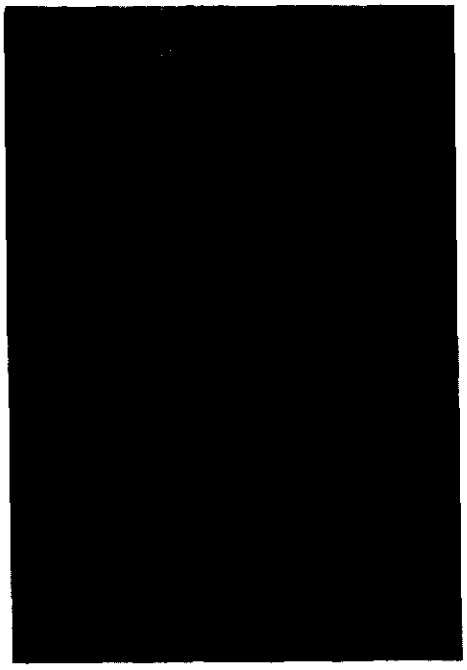

Figure 6. Two large aneurysmal spaces (S) separated by fibrous vascular tissue. Immunocytochemical stain for factor VIII related antigen shows reaction product on the surface of the controlled blood vessel (arrowhead), but not on the surface of the blood-filled spaces (S). (PAP stain, x 100).

(Figure 6). The anti-F VIII RAg, a polyclonal rabbit anti-human antiserum, was obtained from Dakopatts (Copenhagen, Denmark),

DISCUSSION

Despite the fact that many cases of aneurysmal bone cyst have been reported, the specific pathogenesis of this lesion remains uncertain. A number of proposals have been advanced in an attempt to explain the etiology of the lesion. Trauma is often suggested as the causative factor. Bernier and Bhaskar[5] proposed that this lesion was formed as a result of the connective tissue replacement of the hematoma in the bone marrow originating from internal bleeding while keeping communication with the damaged vessels. A vascular theory, proposed by Lichtenstein,[10] suggested that the lesion arose de novo. Due to a vascular change such as an anomalous arteriovenous communication, an elevated venous pressure was produced and eventually dilated and congested vascular spaces were formed. The third hypothesis of the formation of this lesion was proposed by Biesecker et al.[11] who mentioned that the lesion was developed in conjunction with the preexisting primary lesions of bone including fibrous dysplasia,[11-16] ameloblastoma,[17] ossifying fibroma,[18] giant cell granuloma,[11,19,20] hemangioma[11,21] and dentigerous cyst.[22] Then, the primary condition might undergo cystic change and initiate an arteriovenous malformation with the formation of the cystic lesion through the abnormally high hemodynamic forces produced. This mechanism, suggested by Bisecker,[11] might explain the phenomena of cortical bone expansion and the formation of reactive fibro-osseous components and giant cells.

For the present case, we found it difficult to comment on the exact pathogenesis. Although it has frequently, but not for all cases, been reported as a hybrid lesion, no definite primary bone lesion could be identified in the present case. However, findings such as microbleeding and many venous channels within the cellular fibrovascular stroma (Figure 5) as well as the absence of endothelial linings of the aneurysmal spaces as demonstrated by immunohistochemical staining for Factor VIII related antigen, which is a reliable and specific

marker of endothelial cells,[23] seem to support the hypothesis of Bernier and Bhaskar that trauma may be the possible causative factor. But, there is still a question whether trauma can be directly related to its pathogenesis since blows to the jaws are common in the young age group but the occurrence of this lesion is relatively rare in comparison with frequency of trauma upon the jaws.

In conclusion, multiple predisposing factors such as trauma, preexisting bone lesions or other unknown factors appear to be involved in the formation of the aneurysmal bone cyst through an ischemic stromal disintegration with microcyst formation. Then, an abnormal high hemodynamic pressure, resulting, from arteriovenous malformation or damaged vessels might shoot the blood into the coalescing microcysts.

Buraczewski and Dabska[12] proposed the three developmental phases of the aneurysmal bone cyst. The initial phase is predominantly the osteolysis and the cortex is essentially incipient intact. The mid-phase represents the marked bony destruction with the first sign of a "blow-out-appearance." Finally, there is the stabilization phase with the classical radiographic picture of the lesion. Radiographiclly, therefore, the appearance of the lesion is noncharacteristic osteolysis compatible with a large range of diagnosis. In the present case, the stabilization phase was already reached as indicated by the multilocular expansile radiolucency with a thin bony septa giving a soap bubble appearance. However, the rapid growth pattern of the present lesion (within 3 months) and the radiography of a sun-ray appearance may lead to a radiographic interpretation of malignancy.

Microscopically, this cyst should be distinguished from central hemangioma, giant cell granuloma and osteogenic sarcoma especially the telangiectatic type which all share a similar histologic picture.[10] As mentioned above, in the present case, there was no evidence of endothelial lining in the large dilated sinusoidal blood-containing spaces as demonstrated by the immunohistochemical staining for F VIII RAg. Therefore, the central hemangioma is unfavourable. This finding is in accordance with the report of Aho et al.[24] who mentioned that the aneurysmal spaces were devoid of Factor VIII related antigen. On the other hand, circumscribing osteoid spicules surrounding the extremely large blood filled spaces, which is not the usual microscopic picture of the central giant cell granuloma,[25] can be observed in the present case. Thus, the central giant cell granuloma is not suggested. Finally, the finding of the cellular fibrovascular stroma without cellular pleomorphism, abnormal mitosis, and pleomorphic osteoblastic tumor cells can rule out the osteogenic sarcoma.

In view of the treatment modalities, curettage[26,27] with or without bone grafting, is mostly the treatment of choice. En bloc resection[27] and cryotherapy[28] have also been utilized. Radiation, that may induce malignant change,[29] is not recommended unless for those surgically inaccessible cases. Owing to the fairly high recurrent rate as reported by El Deeb et al.[16] and quite a large destructive mandibular lesion, local curettage and cryotherapy of the lesion seemed to be inadequate so that a segmental resection with the bone graft was performed. There was no evidence of recurrence after a close and periodic postoperative follow-up for more than 15 months.

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下顎骨動脈瘤性骨囊腫——病例報告

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動脈瘤性骨囊腫源發於顱顏面骨之病例報告在台灣並不多見。本病例發生於一位十五歲本省籍男性，臨床檢查顯示病人右下顎骨骨體處有一質地堅硬、無痛之腫漲；X光檢查發現右下顎骨骨體處呈骨破壞，並往外擴張腫大；光學顯微鏡檢查發現於疏鬆纖維結締組織中含許多充滿血液之實狀腔及多核巨大細胞，病理診斷爲動脈瘤性骨囊腫。由於此類病竈復發率頗高，且本例病竈侵犯範圍頗廣泛，故採用截段切除術及髂骨移植法之外科治療，術後並做定期追踪檢查，在病理檢查方面本病例應用抗第八因子抗原之免疫組織過氧染色法，發現實狀腔之表面無內皮細胞襯裏，且對第八因子抗原之染色呈陰性反應，此點可與骨內血管瘤有所區別，其結果也有助於瞭解此病變的部分病理機轉。

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Roentgenographic examination showed an expansile osteolytic lesion extruding from the lower border of the right mandibular body with periosteal reaction (Figure 2). Computed tomography revealed an osteolytic lesion with soft tissue density which occupied the right mandibular body extending to the angle area homolaterally (Figure 3).

An incisional biopsy of the lesion under

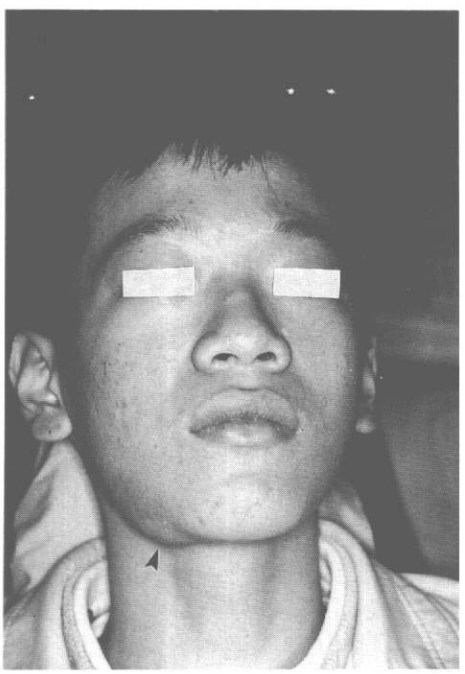

Figure 1. Preoperative view showing a right submandibular swelling (arrowhead).

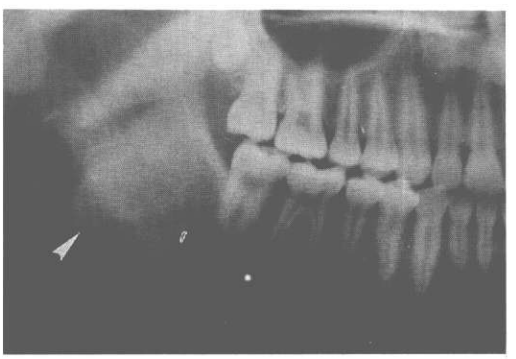

Figure 2. Preoperative panoramic radiograph of right mandible showing an expansile lesion extruding from the lower border of the mandible with periosteal reaction (arrowhead).

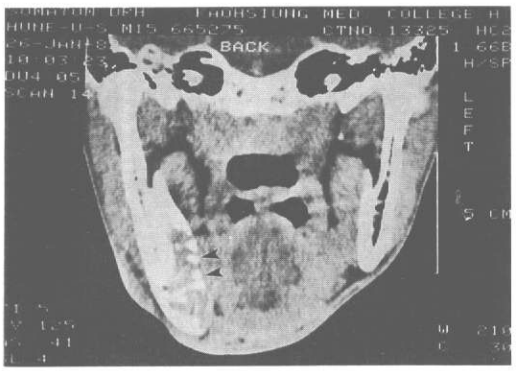

Figure 3. CT scan showing an expansile osteolytic lesion of the right mandibular body (arrowheads).

local anesthesia was performed via an extraoral approach. The right submandibular lymph node was also excised. After removing the paper-thin like expaned cortical bone, many blood filled cystic cavities separated by a thin septa were noticed. Bleeding of a pulsating type was observed and hemostasis was achieved with difficulty. The cavities was packed with a tampon and the wound was closed. The patient was then admitted to our dental ward for the observation of any uncontrollable bleeding after the incisional biopsy and was scheduled for surgery. The pathologic report of the incisional biopsy was consistent for an aneurysmal bone cyst and lymphoid hyperplasia of the right submandibular lymph node. The results of the complete blood and urine examinations were within the normal range with the exception of the alkaline phosphatase which showed an elevated value of 213 IU/1 (normal range: 26.0 – 88.0 IU/1) indicating a possible active osteoblastic and/or osteoclastic activity. Under endotracheal general anaesthesia, segmental resection from the right second mandibular premolar to the ramus of the same side was done. The bone gap resulting from the mandibular resection was bridged by a graft obtained from the right iliac crest which was fixed by stainless-steel miniplates. Following the placement of the bone graft, the submandibular wound was closed in layers. Intermaxillo-mandibular fixation was applied and the occlusion was restored. The postoperative course was uneventful.

Grossly, the resected mandibular specimen measuring about 7 cm in diameter consisted of the right portion of mandible with the associated teeth #29, #30, #31 and revealed a ballooned-out appearance with greatly thinned cortical bone resembling parchment. Upon the removal of the cortical plate, multiple spiderweb-like compartments of varied sizes filled with dark red blood coagulum were observed.

Microscopically, hematoxylin & eosin stained sections showed multiple sinusoidal blood-filled cavities within the fibrous connective tissue in which multinucleated giant cells were scattered and foci of reactive osteoid arranged in a circumscribing distribution were found (Figure 4). Furthermore, small and large venous lakes within a fibrovascular stroma could be observed (Figure 5).

Immunohistochemical staining for Factor VIII related antigen (F VIII RAg), at the concentration of 1:300, by the peroxidase-anti-peroxidase (PAP) method showed that the surface of sinusoidal blood spaces was exclusively devoid of endothelium (Figure 6). However, antibodies to F VIII RAg reacted with their appropriate antigens in the walls of blood vessels which served as internal controls

Figure 4. Microphotograph showing a typical sinusoidal space filled with blood coagulum (left bottom corner) and reactive trabeculae of bones(B) present in the fibrous stroma with scattered multinucleated giant cells (right bottom corner). (H & E, x100)

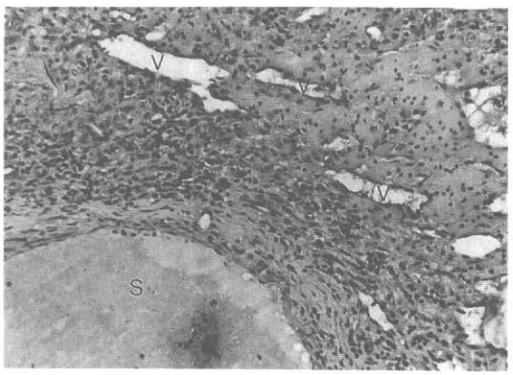

Figure 5. Microphotograph showing many small and large venous lakes (V) within a fibrovascular stroma. A sinusoidal space (S) is at the left bottom corner. (H & E, x100)

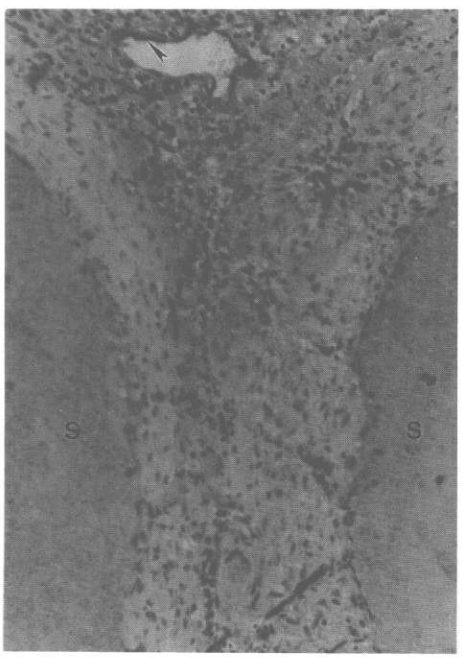

Figure 6. Two large aneurysmal spaces (S) separated by fibrous vascular tissue. Immunocytochemical stain for factor VIII related antigen shows reaction product on the surface of the controlled blood vessel (arrowhead), but not on the surface of the blood-filled spaces (S). (PAP stain, x 100).

(Figure 6). The anti-F VIII RAg, a polyclonal rabbit anti-human antiserum, was obtained from Dakopatts (Copenhagen, Denmark).

DISCUSSION

Despite the fact that many cases of aneurysmal bone cyst have been reported, the specific pathogenesis of this lesion remains uncertain. A number of proposals have been advanced in an attempt to explain the etiology of the lesion. Trauma is often suggested as the causative factor. Bernier and Bhaskar[5] proposed that this lesion was formed as a result of the connective tissue replacement of the hematoma in the bone marrow originating from internal bleeding while keeping communication with the damaged vessels. A vascular theory, proposed by Lichtenstein,[10] suggested that the lesion arose de novo. Due to a vascular change such as an anomalous arteriovenous communication, an elevated venous pressure was produced and eventually dilated and congested vascular spaces were formed. The third hypothesis of the formation of this lesion was proposed by Biesecker et al.[11] who mentioned that the lesion was developed in conjunction with the preexisting primary lesions of bone including fibrous dysplasia,[11-16] ameloblastoma,[17] ossifying fibroma,[18] giant cell granuloma,[11,19,20] hemangioma[11,21] and dentigerous cyst.[22] Then, the primary condition might undergo cystic change and initiate an arteriovenous malformation with the formation of the cystic lesion through the abnormally high hemodynamic forces produced. This mechanism, suggested by Bisecker,[11] might explain the phenomena of cortical bone expansion and the formation of reactive fibro-osseous components and giant cells.

For the present case, we found it difficult to comment on the exact pathogenesis. Although it has frequently, but not for all cases, been reported as a hybrid lesion, no definite primary bone lesion could be identified in the present case. However, findings such as microbleeding and many venous channels within the cellular fibrovascular stroma (Figure 5) as well as the absence of endothelial linings of the aneurysmal spaces as demonstrated by immunohistochemical staining for Factor VIII related antigen, which is a reliable and specific