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Aneurysmal bone cyst of the maxilla. A case report

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INTRODUCTION

The aneurysmal bone cyst (ABC) is a non-neoplastic lesion of the skeleton consisting of a cystic cavity filled with non-endothelial-lined spaces containing blood (Jaffe and Lichtenstein, 1942). Since the ABC was first described in 1942 by Lichtenstein and Jaffe the lesion has been found primarily in the long bones and the vertebral column.

On the other hand, ABC in the maxilla is rare. The first documented case of ABC in the maxilla was presented by Bhaskar et al. (1959). Zachariades et al. (1986), in their review of the literature since that time, found 22 cases with ABC at that site. The etiology and pathogenesis of the lesion have yet to be elucidated. One possible explanation is that the development of the ABC may occur secondarily to a pre-existing bone lesion (Biesecker et al., 1970). We report here a case of ABC coexisted with fibrous dysplasia in the same lesion of the maxilla.

CASE REPORT

On March 28, 1986, a 15-year-old girl with a painless swelling on the left side of the face was referred to our department. The patient stated that the facial swelling had been gradually increasing over a 10-month period.

The past medical history was non-contributory. There was no history of trauma to the left side of the face at any time in the past. On intraoral inspection, a visible dome-shaped protrusion of the soft tissue over the buccal plate of the alveolar bone was seen (Figure 1). It was round, tender, and had a springy consistency. There was a 3 cm swelling in the region of the left first and second premolars and the first molar. A computerized tomography scan of the maxilla permitted confirmation of the destructive, expansive nature of the lesion (Figure 2). On the other hand, left external carotid arteriography revealed hypovascularity throughout the arterial and venous phases.

A mucoperiosteal flap was incised under general anaesthesia, extending from the left maxillary central incisor to the posterior aspect of the left maxillary

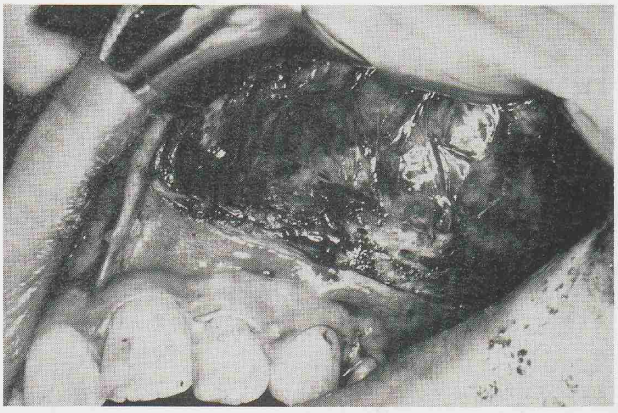

Figure 1. Swelling of the left side of maxilla.

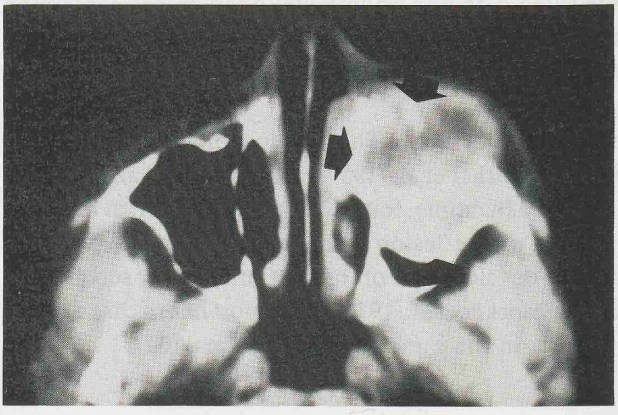

Figure 2. CT-scan of maxilla showing mass (arrow) in anterior wall with expansive invasion.

tuberosity. After reflection of the flap, the thin bony covering over the lesion was then easily removed with a rongeur forceps and the lesion was curretted. Pathologic examination of the specimen revealed an ABC (Figure 3) with fibrous dysplasia (Figure 4). There has been no sign of recurrence in more than three years since the operation.

DISCUSSION

The ABC is a pseudocyst without an epithelial lining, which is filled with numerous erythrocytes. The cavernous cystic walls lined by connective tissue contain hemosiderin and varying numbers of multinuclear giant cells (Jaffe and Lichtenstein, 1942).

The ABC is most commonly associated with younger age populations (Bhaskar et al., 1959). The specific etiology and pathogenesis of the ABC are not fully

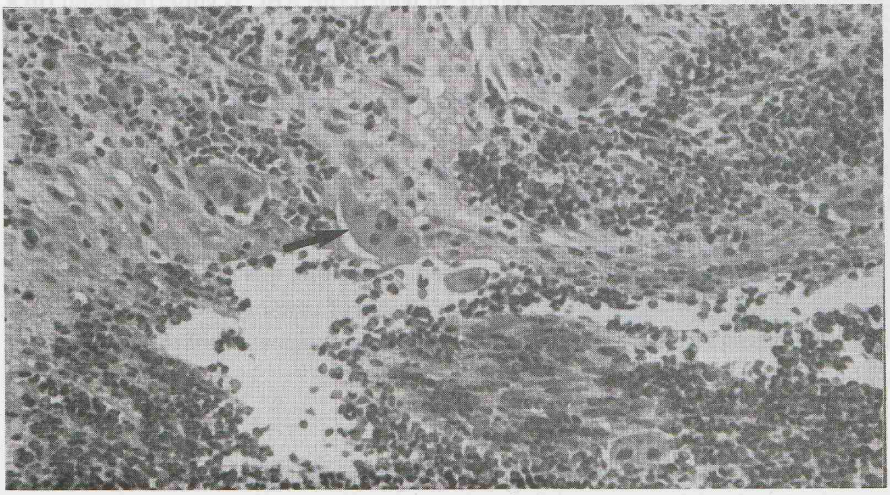

Figure 3. Light micrograph of aneurysmal bone cyst showing a cavernous blood-filled spaces and multinuclear giant cells (arrow). (Hematoxylin-eosin stain, x 200.)

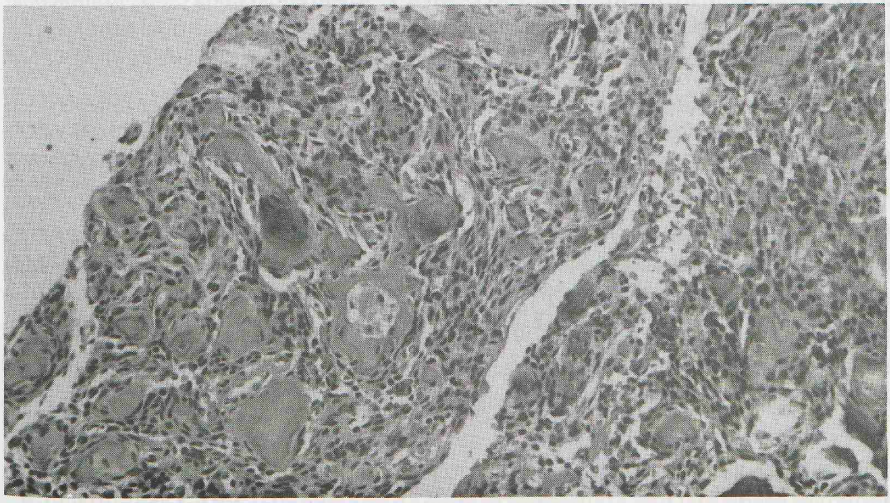

Figure 4. Light micrograph of fibrous dysplasia near aneurysmal bone cyst. Note mesenchymal tissue and bony trabeculae. (Hematoxylin-eosin stain, x 100.)

established, and several theories have been proposed in the literature since ABC was first described:

1. a local circulatory disturbance as a result of trauma (Bernier and Bhaskar, 1985);
2. a local circulatory abnormality (Lichtenstein, 1950);
3. a secondary manifestation of a pre-existent lesion, either in the form of necrosis or a haemorrhagic blow-out (Bieseker et al., 1970).

Secondary ABC is a pathological entity in which the ABC is superimposed on a pre-existing bone lesion. Fibrous dysplasia in the maxilla is a very frequently seen fibro-osseous lesion (Buraczewski and Dabska, 1971). The early lesions in fibrous dysplasia are soft and reddish, because of rich vascularization, and show highly cellular connective tissue, often with mitotic figures and woven immature bone. Also, multinuclear giant cells are seen in the lesion (Bahadur et al., 1986). In our case, it was difficult pathologically to distinguish between primary ABC and spontaneous bleeding into cystic areas of fibrous dysplasia, although the coexistence of fibrous dysplasia and ABC was confirmed. Therefore, it is possible that this case may have developed from a haemorrhagic blow-out of a pre-existing fibrous dysplasia with destruction of the original lesion. A definitive explanation of the etiology and pathogenesis of the ABC remains to be elucidated.

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