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ANEURYSMAL BONE CYST OF THE MAXİLLA REPORT OF A CASE

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SUMMARY: A case of aneurysmal bone cyst of the maxilla is presented and literature reviewed. The term anuerysmal bone tumour is suggested to be more suitable for the condition.

The aneurysmal bone cyst (ABC) is a non-neoplastic, localised, solitary lesion of bone characterised by its replacement with fibro-osseous tissue containing blood filled sinusoidal or cavernous spaces (9,11,16,17).

It is first described as a distinctive pathologic identity by Jaffe and Lichtenstein seperately in 1942. The ABC's had been described under a variety of names viz: hemorrhagic osteomyelitis, ossifying hematoma, osteitis fibrosa cystica, atypic subperiosteal giant cell tumour, aneurysmal giant cell tumour, hemangiomatous bone cyst, subperiosteal bone aneurysm, expansile hemangioma and pulsating benign giant cell tumour (1,3,4,5,11,12,16,17). It affects predominantly the metaphyseal regions of the long bones and the vertebral column. It is a comparatively uncommon bony lesion.

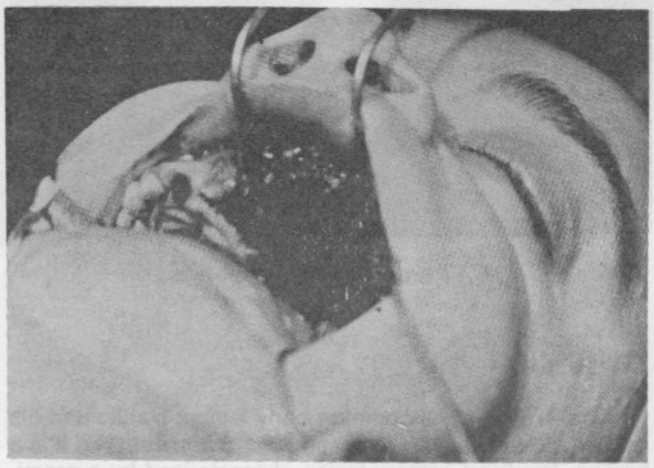
Involvement of the maxilla or mandible is very rare (4,5,7,16,17). Up to date review of the literature revealed 63 reported cases of ABC in facial region. 41 of these were in mandible and 22 in the maxilla (4,17). In this article we present an ABC in the maxilla which we prefer to name as aneurysmal bone tumour because of its agressive behaviour.

CASE REPORT

A 20 year old girl admitted to the Oral Surgery Department of the Dentistry Faculty with complaint of swelling and an insidious pain in her left upper face. Previous treatments, consisting of the extraction of the teeth of the region were of no use. The biopsy of the lesion revealed a diagnosis as an aneurysmal bone tumor and the patient was referred to our clinic, Plastic Surgery Department of İstanbul Medical Faculty for consultation. Physical examination revealed a slightly noticeable assymetry of the face and deviation of the nasal septum to the left side. General examination of the body and organs was non-contributory. Intra-oral examination revealed a firm mass of about 3x4 cm. on the left upper alveolar ridge (Figure 1). The swelling was hard in consistency and the teeth in the area were missing because of previous extractions. There was no sign of inflammation and the overlying mucosa was of normal texture and color. Pressure produced a crepitus sensation and tenderness. There was no palpable cervical lymphadenopathy.

X-ray examination revealed a round, cystic radiolucency extending into the maxillary sinus and cavum nasi, about 4 cm, in diameter. Computerised axial tomography of the region disclosed a destruction beginning in the roots of the extracted upper inciseves, extending to the right side of the alveolus and to the maxillary bone and sinus, destructing the floors of the both sinuses, especially the left side. The nasal cavity was obstructed in both sides, especially in the left-inferior side. The septum was deviated to right. The lesion also produced soft tissue masses on the roof of the oral cavity, as solid densities and was extending to the subcutaneous fatty tissue of the nasolabial region (Figure 2,3,4).

The patient was operated under general endotracheal (oral) anesthesia, beginning with a labio-buccal incision, resecting the invaded alveolar ridge completely and through curretting of both sinuses and nasal cavity (Figure 5). The cavities were packed with gauze iodoforme and incision was closed. The patient had an uneventful recovery and a partial prothesis was to be applied later on. The histopathological report stated: (Macroscopic) The fragments of cortical and spongious bone with reddish-pink coloured soft tissue of about 20 cc. volume were received (Microscopic). Microscopic sections showed that the lesion was composed of sinusoids filled with erithrocytes. The septa around the sinusoids were rich of mononuclear cells, thus forming a thicker wall. Between the mononuclear cells there were occasional osteoclastic giant cells. Around them thin, recently forming bone tissue lamellae were partly observed. Around this, portions of hyalinised connective tissue was observed. In some parts cylindrical ephitelium of sinusal mucosa was seen (Figure 5,6).



Figue 1: Per-operative view of the patient.

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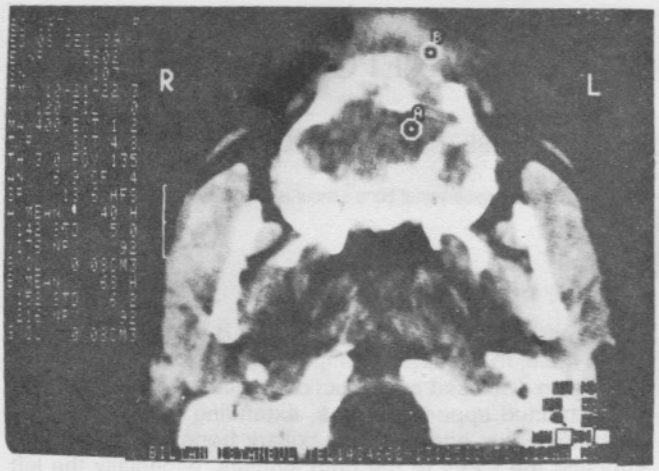
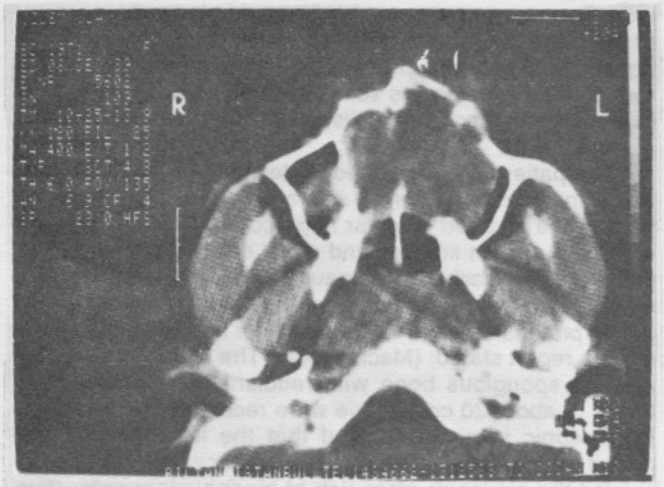




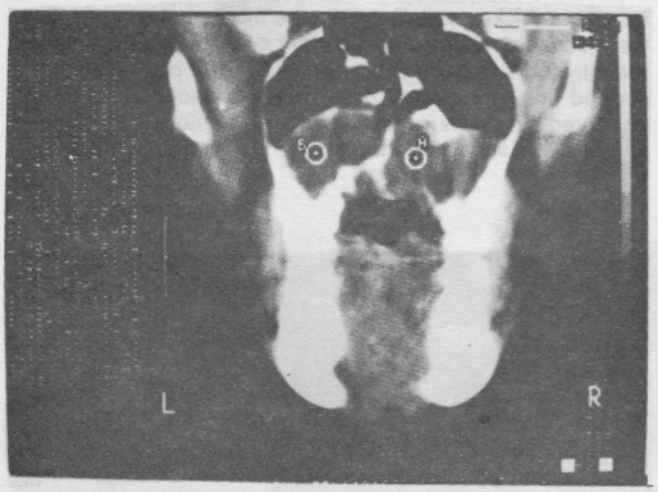
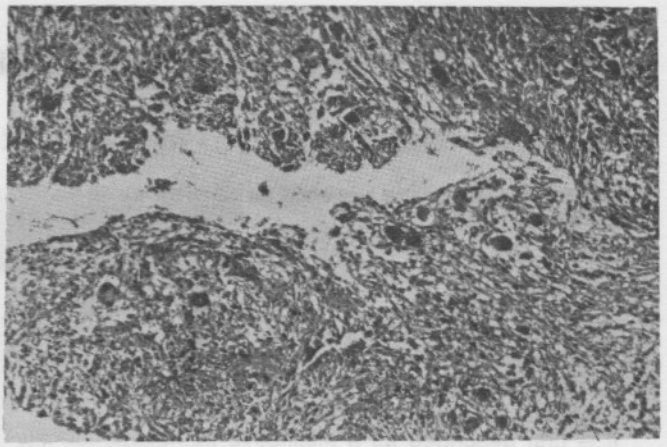


Figure 2, 3, 4: CAT scan views of the patient.



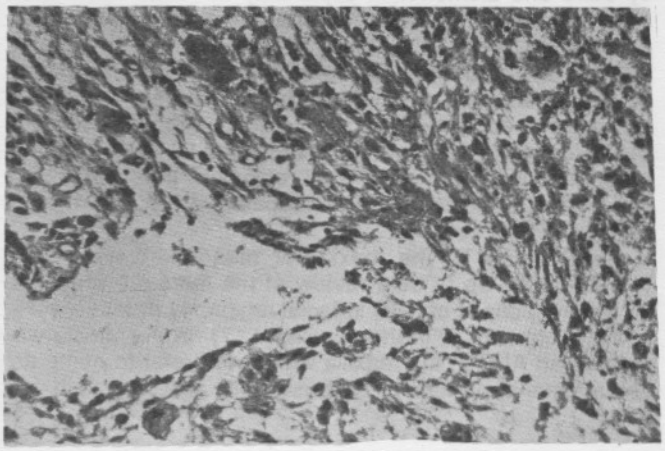


Figure 5, 6: Photographs of the microscopic sections.

DISCUSSION

The ABC is an uncommon bony lesion. Dahlin and his associates reported an incidence of 1.3 percent (16). It is a benign, localised solitary lesion characterised by a prominent bulging of the bone caused by endosteal resorbtion and periosteal apposition of cortical bone due to an underlying fibrous expansion (11). Numereous theories have been proposed regarding the pathogenesis of ABC. Many authors have suggested that the lesion originates as a response to trauma (5,7). Wang reported 71 percent of cases have a history of associated trauma (16). Although a history of trauma may be obtained this is probably secondary related to the expansion of the lesion. In our case there was not any history of trauma. Liechtenstein postulated local circulatory disturbances and altered haemodynamics (6,7). Bernier and Bhaskar argued against a vascular origin and suggested connective tissue replacement of a hematoma (6). However, the vascular origin has been disputed because no endothelial cells or elastic fibers arefound. Biesecker, Marcove, Huvos and Nicke found that 32 percent of cases were associated with different pathological processes of the involved bone, for example fibrous dysplasia (2,6). It has also been reported that it quite frequently arises in association with osteosarcoma, benign osteoblastoma, non-ossifying fibroma, giant cell tumour, benign chondroblastoma, chondromixoid fibroma and giant cell reparative granuloma (2,9,10,12).

According to Thompson (4), the lesion represents a reparative process that follows some undetermined vascular disturbance about the periosteum and surrounding tissues. Possibly the central giant cell granuloma, ABC, traumatic bone cyst are all the same lesion differing only in the stage of development at the time of surgical intervention (3,4).

Buraczewski and Dab ka have distinguished three stages in the development of ABC as an initial stage with prevalent osteolysis and a non-characteristic appearance, a growth phase with marked bony destruction indistinct demarcation and first signs of bony shell and a stabilisation phase with a fully developed radiolucent appearance (4). Radiological features appear as an expansile radiolucent cystic mass that is usually multilocular. In the mandible and maxilla, these features are not as characteristics and a diagnosis connot be made on a radiologic base alone. Trabeculations and a soft tissue mass may be present. The lesion can easily be mistaken for an odontogenic cyst or tumour, a central giant cell reperative granuloma, a lesion of hyperparathyroidism, or other primary and even metastatic lesions (5).

Most of the lesions occur in patients under 20 years of age (% 85) (1,3,4,10,13). Our patient is also 20 years old. The youngest patients reported in the literature is 5 months old (13). The is no significant sex predilection but females have a slightly higher incidence (1,5,13).

Clinically a firm, fainless swelling of the jaw occurs, as it was in our patient. Microscopically the characteristic feature is large blood filled spaces that lack the usual elastic lamine and muscular layers of blood vessels. The walls and septa consist of fibrous tissue containing variable amounts of fibroblasts, benign giant cells and long osteoid trabeculae. Solid zones, when present, may resemble giant cell reparative granuloma and differentitaion may depend solely on the blood filled spaces of ABC (5,12,13).

İn the literature Jacobson was the first to draw attention to the tumoural character of the lesion (8). Tahsinoğlu et al. in their study of 30 cases, suggests that the term aneurysmal bone tumour which is first used by Volkov, is more suitable for the lesion regarding to the characteristics as invasion of the epiphysis, soft tissue mass and periosteal reaction in some cases (13).

Conservative surgical removal is the preferred treatment (1,3,4,5,6,9,11,15,16,17). İrradiation is disputed because it could introduce the possible hazard of irradiation sarcoma. A 53 percent recurrence rate has been reported in the jaws and a 21-59 percent in other bones although the recurrence is considered uncommon by many authors (17).

The rate of recurrence of ABC in the mandible and maxilla is given as 26 percent within the first year (9). The risk of recurrence appeared to be higher in patients younger than 15 years and with lesions smaller than 5 cm. Since this is a high recurrence rate patients should have frequent follow-up examinations. If the lesions recur, conservative surgical treatment is again indicated with the possibility of hyperparathyroidism excluded. We have not met any recurrence during the follow-up period of one year.

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