Plexiform Ameloblastoma

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Abstract
Ameloblastoma, a benign epithelial odontogenic tumor, is locally aggressive. This tumor comprises about 1% of tumors and cysts arising in the jaws. It appears most commonly in the third to fifth decades and with equal frequency between sexes. Ameloblastoma prevalently occurs in the mandibular molar and the ramus areas. Recurrence frequently appears after inadequate treatment. They are usually benign in growth pattern but frequently invade locally and occasionally metastasize. In the present study, a case of a large plexiform ameloblastoma was presented with its clinical, radiological, histological features and treatment modalities.

Key words: **Ameloblastoma, Mandible, Odontogenic Tumors**

Özet
Selim epitelyal odontojenik bir tümör olan ameloblastom bölgesel olarak agresiftir. Bu tümör çenelerde oluşan kist ve tümörlerin yaklaşık %1'ini kapsar. Daha yaygın olarak hayatın üçüncü veya beşinci dekatlarında ve her iki cinste eşit sıklıkta görülür. Ameloblastom sıklıkla mandibular molar ve ramus bölgelerinde oluşur. Rekürens ekseriyetle yetersiz tedavi sonrasında görülür. Genellikle selim bir büyüme şekli vardır ancak yerel olarak invaze olur ve nadiren metastaz yapar. Bu çalışmada, büyük bir pleksiform ameloblastom vakası klinik, radyolojik, histolojik özellikleri ve tedavi yöntemleri ile sunuldu.

Anahtar Kelimeler: **Ameloblastoma; Mandibula, Odontojenik Tümörler**

Submitted : March 05, 2007
Revised : July 19, 2008
Accepted : April 27, 2009

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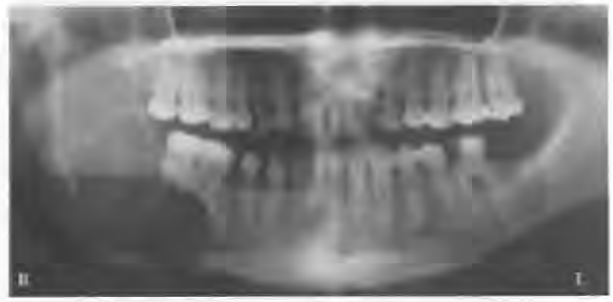
Introduction

Ameloblastoma is a true neoplasm of odontogenic epithelium (1). It represents about 1% of all oral ectodermal tumors and 9% of odontogenic tumors (2). It is an aggressive neoplasm that arises from remnants of the dental lamina and dental organ (odontogenic epithelium) (3). Most ameloblastomas develop in the molar-ramus region of the mandible with 70% of these arising in the molar-ramus area and they are occasionally associated with unerupted third molar teeth (4). Ameloblastoma appears most commonly in the third to fifth decades but the lesion can be found in any age group including children (2, 5, 6). Its histological appearance is similar to that of the early cap-stage ameloblastic elements of developing without complete differentiation to stage of enamel formation (1, 6). Six histological subtypes of ameloblastoma have been identified and comprise follicular, plexiform, acanthomatous, granular, basal cell and desmoplastic type (5). It is well known that ameloblastoma can be radiologically unilocular or multilocular radiolucency with a honeycomb or soap bubble appearance (5). Even it frequently recurs after inadequate surgical treatment (7), ameloblastomas infrequently metastasize (8). The plexiform-unicystic pattern is less aggressive and has a significantly lower recurrence rate (9). The most common sites of metastases are the lungs followed by regional lymph nodes, pleura, vertebrae, skull, diaphragm, liver, parotid and small intestine (8). Mechanisms of distant spread are debated and include aspiration, haematogenous spread, lymphatic spread and malignant activation of the developmental rests of epithelial tissue (8).

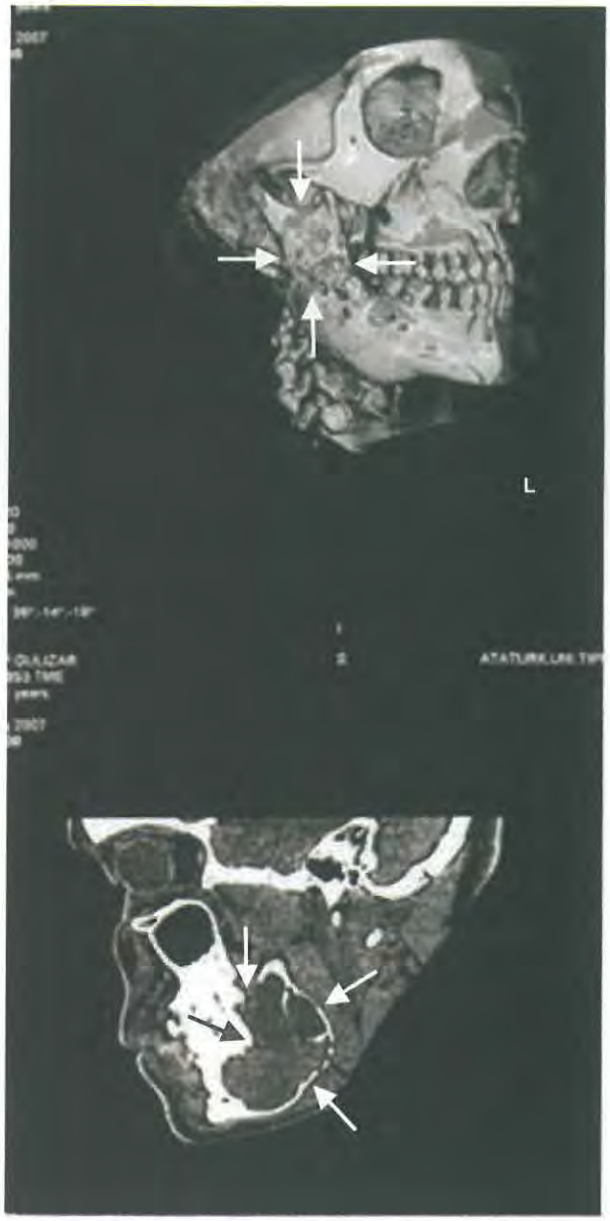
Case report

A 27-year-old woman had been applied to Department of Oral Diagnosis and Radiology of the School of Dentistry of Ataturk University with complaint of expansion on the right mandibular molar region. The medical history was unremarkable. Clinical examination revealed a diffuse, smooth-surfaced and hard swelling on the ascending ramus and molars region of the mandible. The swelling was large, expansive, and painless. It was covered by normal mucosa.

Panoramic radiography showed a large multilocular radiolucent area (about 50 by 87 mm) occupying the right mandible from the first molar tooth to the neck of condylar process and the coronoid process including the right ascending ramus area (Pic. 1). Expansion of the lesion had caused displacement of the mandibular canal. Root resorption was observed in the first and second molar. The base of the mandible and the anterior border of the ramus was damaged and thinned. Carious was observed in the second molar. In order to determine the lytic lesion in more detail, Computerized Tomography (CT) was taken. Sagittal CT showed an expansive lesion, erosion, cortical destruction and thinning (Pic. 2).

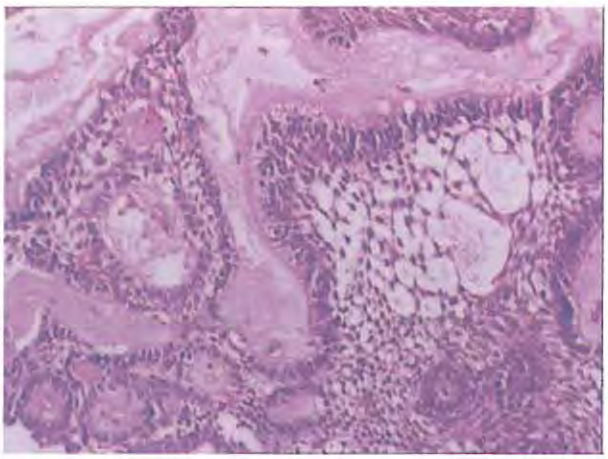


Picture 1. Panoramic radiogram revealed a large multilocular radiolucent area (limited with white arrows), about 50 by 87 mm, extending from the right first molar to the right coronoid process including the right ascending ramus area.



Picture 2. Sagittal CT showed a large (limited with white arrows), expansive mass occupying the right mandible from the condyle to the right first molar tooth. An expansive lesion, erosion, cortical destruction and thinning was also observed.

The histopathological processing of the tumor revealed a plexiform ameloblastoma predominantly composed of epithelium arranged as a tangled network of anastomosing strands enclosing cysts of various size (Pic. 3).



Picture 3. The histopathological processing of the tumor revealed a plexiform ameloblastoma predominantly composed of epithelium arranged as a tangled network of anastomosing strands enclosing cysts of various sizes (H/E X200).

Discussion

Ameloblastoma is a benign epithelial odontogenic tumor but is often aggressive and destructive, with the capacity to attain great size, erode bone and invade adjacent structures (7, 10). Although the term 'ameloblastoma' was coined by Churchill in 1933, the first detailed description of this lesion was by Falkson in 1879 (8). It is the most common odontogenic tumor although it represents only about 1% of tumors and cysts of the jaws (5).

Twenty percent of ameloblastomas occur in the maxilla and they are localized most often in the canine and antral regions of upper jaw. In the mandible (80% of ameloblastomas), 70% are located in the area of the molars or the ascending ramus, 20% in the premolar region, and 10% in the anterior region (2, 11). However, it rarely affects the soft tissue (peripheral ameloblastoma) (5). About 10-15% of ameloblastomas are associated with a non-erupted tooth (11). In the present case, a large plexiform ameloblastoma found the ascending ramus and molars region of the mandible and it was not associated with a non-erupted tooth.

It appears with greater frequency in the third or fourth decade of life, except in the case of the unicystic variety, which is diagnosed between the ages of 20 and 30 years although cases have been described in children (2, 4, 6). Ameloblastoma appears equal frequency between sexes (11), although a higher frequency in females than in males has been described (8). In our case, the patient was female and was second decade of her life.

Ameloblastoma generally occurs in bone. It is thought to originate from sources that include cell rests of the enamel organ; epithelium of odontogenic cysts; disturbances of the developing enamel organ; basal cells of the surface epithelium or heterotrophic epithelium in other parts of the body (3, 6, 12). Several causative factors have been proposed, including nonspecific irritating factors such as extraction, caries, trauma, infection, inflammation, or tooth eruption; nutritional deficit disorders; and viral pathogenesis (9). In the present case, we thought ameloblastoma to originate from source that nonspecific irritating factor such as carious placed the first molar tooth of the right mandible.

Clinically, it frequently manifests as a painless swelling, which can be accompanied by facial deformity, malocclusion, and loss of dental pieces, ulceration and periodontal disease and paresthesia of the affected area (6, 11). However, it is still infiltrative in character in terms of local malignant behavior (2). In our case, clinical examination revealed a large, expansive mass in the ascending ramus and molars region of the mandible. The swelling was hard, painless to palpation and covered by normal mucosa.

In most cases ameloblastoma present a characteristic but not diagnostic radiographic appearance (8). Ameloblastoma may present in three different patterns. The most common form is the multilocular with various cysts that are in groups or separated by osseous reinforced septa (soap bubble appearance). Another image is a beehive pattern, this being the second most common type. A third radiographic manifestation, which is very important in terms of a differential diagnosis, is the unilocular form (2). Resorption of the adjacent tooth roots is not uncommon (8). In many cases an unerupted tooth, most often a mandibular third molar, is associated with the tumor (12). Sometimes ameloblastoma is indistinguishable from a dentigerous cyst (8).

Ameloblastoma has a persistent and slow growth, spreading into marrow spaces with pseudopods without concomitant resorption of the trabecular bone. As a result, the margins of the tumor are not clearly evident radiographically or grossly during operation, and the lesion frequently recurs after inadequate surgical removal, showing a locally malignant pattern (8). Panoramic radiography is simple and inexpensive method, which can be used in daily practice (13). The structure of such lesions can be detected on panoramic radiographs. Therefore, panoramic radiographs may be preferred before CT. However panoramic radiographs are inadequate to localize such lesions because of the nature of the panoramic radiography, with its inherently less-sharp image, and ghost image (1). Hence, CT is usually helpful in determining the contours of the lesion, its contents and its extension into soft tissues for diagnosis (14). Ameloblastoma typically shows expansive growth with an osseous shell. CT findings include cystic areas of low attenuation with isoattenuating solid regions and Contrast-enhanced CT shows an enhancement effect in the solid components (4). Although there are no appreciable differences between MRI and CT for detecting the cystic component of the tumor, MRI is slightly superior (15, 16).

Histologically, ameloblastoma is characterized by the proliferation of epithelial cells arranged on a stroma of conjunctive vascular tissue in locally invading structures that resemble the enamel organ at different stages of differentiation (6). Diverse histological patterns have been described in the literature, including those with follicular, plexiform, acanthomatous, papilliferous-keratotic, desmoplastic, of granular cells, vascular and with dentinoid induction (5, 6, 8). The tumor found in our patient was an ameloblastoma of the plexiform type. The term "plexiform" refers to the appearance of anastomosing islands of odontogenic epithelium in contrast to a follicular pattern (17).

Ameloblastoma is divided into three clinicoradiologic groups: solid or multicystic, unicystic and peripheral. Solid ameloblastoma is the most common form of the lesion (86%). It has a tendency to be more aggressive than the other types and has a higher incidence of recurrence (12). Unicystic ameloblastoma has a large cystic cavity with luminal, intraluminal or mural proliferation of ameloblastic cells. It is a less aggressive variant and it has a low rate of recurrence (9, 12, 18),

although lesions showing mural invasion are an exception and should be treated more aggressively (18). Peripheral ameloblastoma exists in soft tissue. The observation of Ueno et al.(19) and El-Mofty et al.(20) that most peripheral ameloblastomas are plexiform. Based on clinicoradiologic feature of the tumor in our case, the tumor was a multicystic ameloblastoma.

Treatment of mandibular ameloblastoma continues to be controversial. Because of their neoplastic nature, surgical treatments differ from those of other cystic lesions. Prior to choosing a treatment for ameloblastomas, the clinicoradiologic variant (solid, multicystic, unicystic, peripheral), anatomic location, clinical behavior and size of the tumor, and age of the patient should be assessed (21). Besides surgery, treatment may also include cryo-, radio- and chemotherapy (22, 23). Ameloblastoma is usually resected enbloc and sometimes with hemimandibulectomy or partial maxillectomy if the lesion is highly infiltrative and extensive. Therefore, precise preoperative diagnosis has important therapeutic implications (4). Rates of recurrence may be as high as 15% to 25% after radical treatment and 75% to 90% after conservative treatment (23). Surgical resection with margins of 1–2 cm has had the least rate of recurrence, in spite of the variant (2). When treated inadequately, malignant development is a possibility (24). Long - term follow-up is necessary because this lesion has been shown to recur 25 and 30 years following primary treatment (8, 22). When treated inadequately, malignant development is a possibility (24). Metastatic dissemination in ameloblastoma is rare but it does occur (8).

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