

Follicular Ameloblastoma

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Abstract: Ameloblastomas also known as adamantinoma are slow-growing, locally invasive tumour affecting the maxillofacial region. There are various histologic variants of ameloblastoma; most common of it is follicular ameloblastoma. Follicular ameloblastoma is commonly seen in elder people, with high recurrence rate. Radiographic finding usually shows a multilocular radiolucency with well-defined borders. This paper presents a case of follicular ameloblastoma in a 55-year-old male who had a swelling on the right mandible region.

Keywords: Ameloblastoma, follicular, mandible, multilocular, recurrence

1. Introduction

The term odontogenic tumour defines many lesions including hamartomatous or benign tissue proliferation to malignant neoplasm with metastatic potential. They are derived from epithelial, ectomesenchymal and/or mesenchymal elements and are found entirely within maxillofacial skeleton (intraosseous/centrally located) or in the soft tissue (gingiva) overlying tooth-bearing areas or alveolar mucosa in edentulous regions (extraosseous or peripherally located)¹. Etiology of benign and malignant odontogenic tumours is unknown and most of the odontogenic tumours appear to arise without an obvious causative factor².

Ameloblastoma is one of the most commonly occurring odontogenic tumours of jaws, represents one percent of all tumours of head and neck region³. WHO in 2005 clinically classified ameloblastoma into (a) solid/multicystic, (b) unicystic, (c) extraosseous/peripheral and (d) desmoplastic. There are two basic histopathologic patterns in solid/multicystic ameloblastoma: (1) follicular and (2) plexiform. Other microscopic patterns of ameloblastoma include acanthomatous, basal cell-like and granular cell. These patterns can be uniform or mixed. In different parts of the world, the distribution of ameloblastoma varies to a certain extent⁴. Histologically, the tumour is composed of odontogenic epithelium arranged in islands or anastomosing cords separated by connective tissue. The former growth pattern is referred to as follicular whereas the latter is called plexiform. The 2 growth patterns may coexist within the same tumour⁵.

2. Case Report

A 55-year-old male patient reported with a chief complaint of swelling in the lower right back tooth region for 3 months.

History of present illness revealed that the patient was apparently asymptomatic 3 months back when a swelling appeared in the mandibular right posterior region. Initially, the swelling was small in size, but it gradually increased to the present size. The swelling was not associated with pain. No history of trauma and pus discharge from that region was noted. Past dental, medical and family histories were non-contributory. On general physical examination patient was moderately built and nourished. On extraoral examination a diffused swelling was noted on the right facial region. Overlying surface of swelling was smooth and of normal

skin colour. On palpation, it was nontender, soft in consistency, with no local rise in temperature. The right submandibular lymph node was palpable and nontender.

The intraoral examination, revealed a sessile, solitary, well-defined, oval swelling in relation to right labial mucosa, measuring about 0.5 cm x 0.5 cm in diameter extending from 44 to 47 teeth region. Mucosa over the swelling appeared pale pink. There was vestibular obliteration with respect to 44, 45 and 46, regions. (figure 1)



On palpation, swelling was well defined, non-tender, soft to firm in consistency with no associated discharge. Slight mobility was present with respect to 45 and 46.

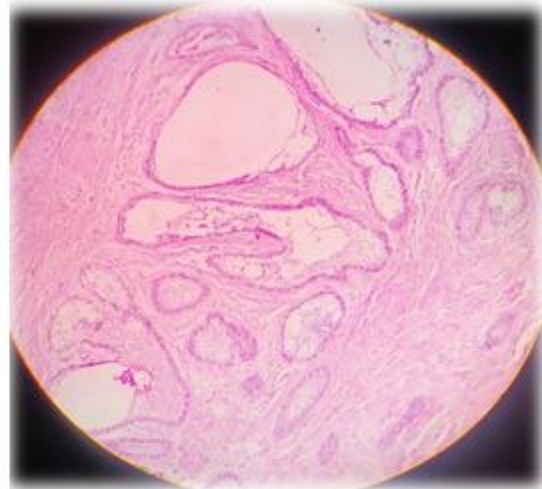
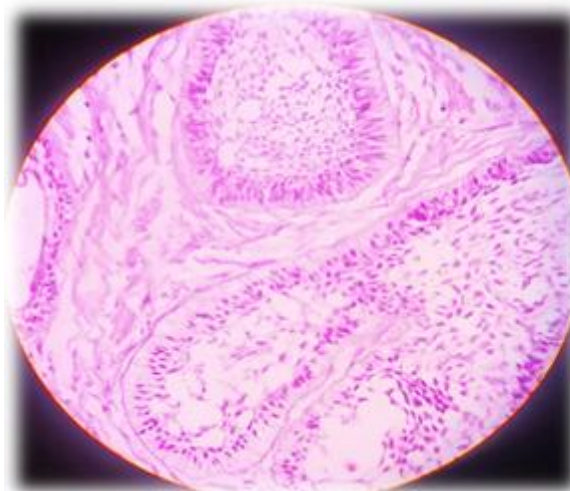
Investigations such as orthopantomogram (OPG), blood investigations, and histopathology examination were done. Blood investigations were found to be within normal limits. OPG showed a well-defined radiolucent lesion in the mandibular posterior region, extending from 44 to 46 regions. Root resorption in relation to 45 was noted. Internal structure appears to be radiolucent with radiopaque internal septa giving a multilocular appearance (figure 2).



On the basis of history, clinical and radiographic examinations, a provisional diagnosis of odontogenic cysts or ameloblastoma in the right mandibular region was given. Differential diagnosis of odontogenic myxoma and central giant cell granuloma (CGCG) was considered.

The lesion was excised using diode laser in dept of periodontology with fiber-optic tip at 1.5 W in a contact, continuous wave mode. Excised specimen was sent for histological examination.

In the department of oral and maxillofacial pathology, we received a single bit of soft tissue, firm in consistency, yellowish white in colour, measuring approx. 0.9x0.5cm in diameter. The H and E stained section showed a proliferating odontogenic epithelium in the form of follicles. Peripheral cells in the follicles were tall columnar and the central part of follicle showed stellate reticulum like tissue with microcysts. The connective tissue stroma was dense and fibrosed. Higher magnification, showed tall columnar ameloblast like cells with polarised nuclei arranged in follicles, with central region consisting of loosely arranged cells resembling stellate reticulum. Histological report was suggestive of follicular ameloblastoma. So final diagnosis of follicular ameloblastoma was given. (Figure 4 and 5)



3. Discussion

Ameloblastoma has a known history of 190 years. During different periods, it has been known by different terms. Gorlin identifies Cusack as the first person to identify "Ameloblastoma" in 1827 and Falkson gave a full report and description of ameloblastoma in 1879. Malassez (1885) introduced the term "Adamantine epithelioma" while Derjinsky (1890) introduced the term "Adamantinoma". Ivy and Churchill, in 1930, used the term "Ameloblastoma" which is the preferred terminology till date¹. Robinson defined ameloblastoma as "unicystic, non-functional, intermittent in growth, anatomically benign and clinically persistent tumour." It may arise from residual epithelium of tooth germ, odontogenic cysts or enamel organ³. Grossly, ameloblastomas are composed of one or more cysts filled

with gelatinous material or clear to straw-colored fluid⁵. Ameloblastomas account for 1% of benign tumours and cysts of the jaw. Incidence is estimated to be 0.5 cases per million person-years worldwide⁶. Robinson on reviewing 293 cases reported site incidence of 83.7% in the mandible and 16.3% in the maxilla⁷. Multicystic/solid is the most common form of ameloblastoma. They can present with huge swellings over the jaws which can result in disturbances in facial aesthetics and function, such as difficulty with mouth opening, swallowing, chewing, breathing, neurologic deficits, and pathologic fractures. Up to 80% of ameloblastoma cases occur in the mandible, with a predilection for the posterior mandibular region^{7,8}. Reichart and coworkers reviewed 3677 cases of ameloblastoma reported in the literature between 1960 and 1993. The median age at presentation was 35 years (range, 4–92 years)

and the male to female ratio was 1.14:1.0. Eighty percent of ameloblastomas occurred in mandible, around the molars and ramus area⁹. The present case was seen on mandible region of a male patient but not in the molar ramus region. Typically, ameloblastoma present as painless slow growing mass as in our case, it took about three months for the swelling to reach the present size. Normal functions likeswallowing, chewing and breathing, was not severely impaired in our case

Radiographically ameloblastoma usually show as an expansile, radiolucent, multiloculated cystic lesion, with a characteristic “soap bubble-like” appearance. We can also find a unilocular radiolucent lesion, there can be thinning and expansion of the cortical plate with erosion and displacement and resorption of adjacent teeth⁹. In the present case, a well-defined radiolucent lesion was seen in the mandibular posterior region with root resorption in relation to 45, which indicates a destructive growth. Internal structure appeared to be radiolucent with radiopaque internal septa giving a multilocular appearance, which is common of ameloblastic radiographic features.

There are many histopathological subtypes of ameloblastoma-follicular, plexiform, acanthomatous, desmoplastic, granular cell, and basal cell pattern, that may exist singly or as a combination of two or more types¹⁰. Follicular and plexiform are the commonly encountered variants accounting for 32.5% and 28.2% respectively; followed by the acanthomatous subtype 12.1% and desmoplastic 4-13%. Follicular ameloblastoma consists of discrete follicles with resemblance to the stellate reticulum of enamel organ, with variable amount of tissue stroma. Follicular subtype of ameloblastoma is the most common variant, so some pathologists believe that the acanthomatous, granular cell, basal cell, and desmoplastic variants are subsets of the follicular ameloblastoma¹¹. Our case showed a proliferating odontogenic epithelium in the form of follicles. Peripheral cells in the follicles were tall columnar ameloblast like cells with polarised nuclei and the central part of follicle showed stellate reticulum like tissue but with microcysts. The connective tissue stroma was dense and fibrosed with no continuous line of anastomosis.

Most common differential diagnosis of ameloblastoma involving the mandibular posterior region are odontogenic keratocyst, CGCG, odontogenic myxoma, primary hyperparathyroidism, secondary hyperparathyroidism, central haemangioma, and aneurismal bone cyst. Surgery is the standard treatment for ameloblastomas. Data from 82 ameloblastoma specimens showed microscopic tumour extension 2–8 mm (mean of 4.5 mm) beyond the radiographic boundaries of the tumour. Hence recommended bone margins are 1–1.5 cm for unicystic and 1.5–2 cm for solid/multicystic histological types, and provides increased cure rates^{12,13}. Complete surgical remove of the lesion was done and follow up of the case for eight months, has not reported recurrence.

According to the WHO in 1992, ameloblastoma is a benign but locally invasive polymorphic neoplasm consisting of proliferating odontogenic epithelium, which usually has a follicular or plexiform pattern, lying in a fibrous stroma (1).

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4. Conclusion

Ameloblastoma has got lot of variants based on histologic patterns. Histologic variant of ameloblastoma should be diagnosed properly and treated accordingly. Long-term follow-up is required in ameloblastoma to check for recurrence. Ameloblastoma should be treated immediately when diagnosed because long-standing cases may cause facial disfigurement. The prognosis of the treatment is basically dependent to the extension of the lesion and adjacent structures involvement rather than origin of lesion. The need for a definite treatment protocol and lifetime periodic follow-up for detection of recurrence is needed as even a five year tumour-free period does not necessarily mean a cure.

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