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CASE REPORT **OPEN ACCESS**

MANDIBULAR FOLLICULAR AMELOBLASTOMA IN AN ELDERLY PATIENT–A CASE REPORT

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ABSTRACT

Ameloblastoma is a benign locally invasive epithelial odontogenic tumour comprising 1% of all tumours and cysts arising in the jaws. It is commonly found in the third and fourth decade in the molar ramus region of the mandible. The commonest histopathological variant of this disease process is follicular type having the highest recurrence rate. Ameloblastomas frequently occur in relatively young people, but are rarely seen in people aged 70 years or older.A case of follicular ameloblastoma of mandible in a 72 years old male is discussed here with detail clinical, radiological, and histopathological features.

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INTRODUCTION

Ameloblastoma is a tumor originating from the epithelium involved with formation of the teeth. It has aggressive behaviour and recurrent course, but rarely metastatic.This disease process represents 1% of all tumors and cysts that involve the maxillomandibular area and about 10% of all odontogenic tumors (1,2). The term ameloblastoma was suggested by Ivy and Churchill in 1934 based on odontogenic epithelial etiology.However, it was first reportedmuch earlier by Broca, in 1868.

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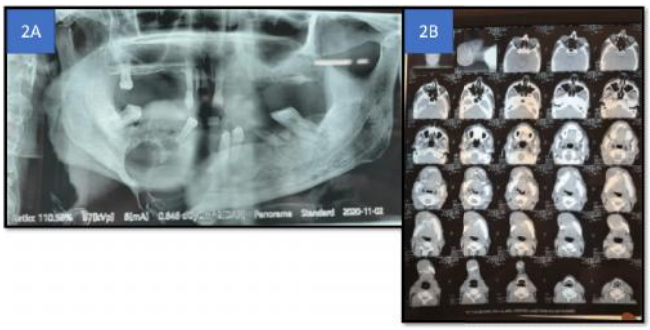
About 80% of ameloblastoma are found in lower jaw, and less frequently on maxilla. The age group predilection peaks in the 20s and 30s, with the average age being between 30 and 40 years, and the majority of cases occur in the 30 to 60 years age group (3-6) Based on these figures, ameloblastomas are considered to be fairly rare in the elderly.

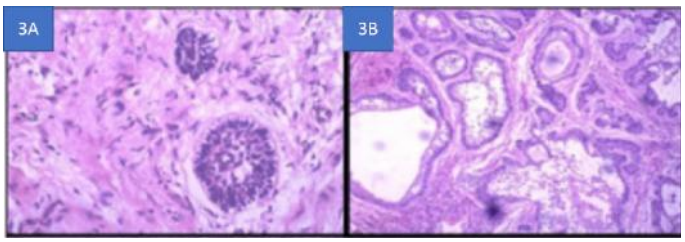
CASE REPORT

A 72 year old male patient reported to the Department of Oral & Maxillofacial Pathology, Guru Nanak Institute of Dental Sciences and Research, Panihati, Kolkata with a chief complaint of swelling involving right side of face since last few months. The swelling was initially small & asymptomatic which gradually increased in size.

Figure 1. A,B,C – Extra oral profile view , 1D – Intra oral view



2A – Orthopantomogram (OPG) of the patient, 2B – CBCT images



3A- H & E (10x) showing histopathology of incision biopsy, 3B)H& E (10x) showing follicular pattern with areas of cystic degeneration

Clinical examination revealed the presence of extraoral swelling involving the body of the mandible on the right side measuring 8 cm X 5 cm. On palpation, the swelling was slightly tender, firm to hard in consistency with fixation of the overlying skin. Regional lymphadenopathy, local rise of temperature and fluctuation were also evident, however, parasthesia/anesthesia of the lower lip and reduced mouth opening was not elicited. Intraoral findings showed partially edentulous upper and lower jaws with a relatively diffuse, large, firm to hard, round to ovoid, fluctuant, non-compressible swelling measuring about 10cm x 8cm, extending from the lingual surface of the alveolar ridge to the buccal vestibule labiolingually while the mesiodistal extension was from the distal aspect of 44 uptothe mesial aspect of 48.Marked expansion of buccal and lingual cortical plates was recorded in association with egg-shell crackling especially on the buccal side. The overlying mucosa was discoloured with detectable white patches. The tongue movement was normal. Ortho pantomogram (OPG) revealed large, relatively well-defined, multilocular radiolucency characterized by typical soap-bubble appearance with scalloped borders, involving the entire body of the mandible, on the right side. Expansion and thinning of the inferior border leading to its discontinuation was also noted. Preoperative routine haematological and biochemical investigations were within normal limits. Based on the clinical and radiological findings, the patient was referred to the Department of Oral & Maxillofacial Surgery for further treatment & management with a provisional diagnosis of odontogenic cyst or neoplasm.

Incisionalbiopsy was performed and the specimen was evaluated later on. Section stained with H/E revealed the presence of actively proliferating neoplastic odontogenic epithelial cell arranged in follicles bounded peripherally by tall columnar ameloblast like cells with centrally placed cells simulating stellate reticulum, in a fibroblascular connective tissue stroma showing variable inflammatory response. Few islands revealed hypercellularity and increased mitotic activity at places and extensive epithelioid differentiation leading to loss of stellate reticulum phenotype. Areas of necrosis could also be noted.Overall light microscopic features are suggestive of “Follicular Ameloblastoma”. To rule out the malignant transformation Immuno histochemistry was performed which revealed positivity for CK-18, and focal faint expression of proliferation marker (KI 67). Hence, the confirmatory diagnosis was that of “Follicular Ameloblastoma.” – benign type. With the confirmatory diagnosis, the patient was referred to the Department of Oral & Maxillofacial Surgery for further treatment & management.

DISCUSSION

Literature shows that although ameloblastoma is usually a benign odontogenic tumor, representing only 1% of all tumors and cysts of maxilla and mandiblebut it is highly recurrent and rarely metastatic.(7)The majority of patients with this disease process are asymptomatic,however, tumoral expansion may lead to development of symptoms. In our case the patient was also asymptomatic initially but swelling developed later on. In explanation of etiology of Ameloblastoma, Csiba., et al. proposed (10) various etiological factors like exodontias, caries, trauma, infections, inflammations or dental eruption; diseases caused by nutritional deficiencies and viral pathogenesisare also to be considered. In the case reported here, the patient also showed chronic gingivitis, which was in accordance to the proposition made by Csiba et al (8-10). According to Mendenhall et al, patients may present with a slow-growing mass, malocclusion, loose teeth, or more rarely paresthesia and pain; however, many asymptomatic lesions are detected incidentally on radiographic studies. (5) Slow growing mass with malocclusion was the chief complain of our patient. Radiographically, ameloblastoma of the mandible can mimic other tumors of the mandible, such as, the odontogenic keratocyst, aneurysmal bone cyst, central giant cell tumor. (11) So, its final diagnosis can only be confirmed through a histopathological examination. Based on the clinical and radiological findings, a provisional diagnosis of odontogenic cyst or neoplasm was made in our case. Gardner DG stated that diverse histological patterns have been described in the literature and which include follicular, plexiform, acanthomatous, papilliferous-keratotic, desmoplastic, granular, vascular and those with dentinoid induction. (11) It is generally accepted that there is no relationship between the individual patterns and the behaviour of the tumor or its prognosis. (12) The tumor found in our patient was that of an ameloblastoma of follicular type. The term “follicular” refers to the appearance oftumor islands consisting of neoplastic, odontogenic epithelial cells resembling ameloblasts surrounding the stellate reticulum-like cells, and arranged in follicles with variable stromal inflammatory response. Several types of odontogenic lesions, particularly odontogenic keratocyst and solid ameloblastoma, although defined as benign, demonstrate locally aggressive behavior and a

potential lethal nature. Immunohistochemical assessment in between different histological types of ameloblastomas will help in predicting their aggressive biological behavior. Immuno-histochemistry was also performed which revealed positivity for CK-18, and focal faint expression of proliferation marker (KI 67). The mainstay of treatment is surgery, wide resection recommended due to the high recurrence rate of solid/multicysticameloblastomas. The recurrence rate after resection is 13–15%, as opposed to 90–100% after curettage. Many authors recommend a margin of 1.5–2 cm beyond the radiological limit to ensure all microcysts are removed. Several studies suggest treatment as an important prognostic factor, specifically implicating undertreatment as a cause of recurrence. While not a first line treatment, radiotherapy should be considered for patients with positive margins who are not amenable to re-excision. Follow-up is essential as most recurrences present within the first 5 years; however, some have been observed for more than 10 years after initial treatment (9-13).

CONCLUSION

Ameloblastoma is one of the most commonly occurring benign tumor of odontogenic epithelial origin involving mandibular body-ramus region affecting middle aged group, yet other differential diagnoses such as odontogenic keratocyst, odontogenic myxoma, central giant cell granuloma have to be excluded with advanced radiological and histopathological techniques. As facial deformity poses a frequent problem, early diagnosis along with conservative surgical intervention should be the treatment of choice. In conclusion, although ameloblastoma is one of the most common odontogenic tumors, an early diagnosis and proper treatment can improve the quality of life of an individual.

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