AMETOBLASTIC FIBROMA- An unusual jaw tumor

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Abstract :
Ameloblastic fibroma is a rare benign tumour which, according to WHO classification, belongs to the class of mixed odontogenic tumours. It presents as a uni or multilocular radiolucent lesion without cortical breach. Two forms of ameloblastic fibroma are postulated a true neoplasm with recurrent malignant potential and a hamartomatous lesion occurring in childhood. We present a rare case of ameloblastic fibroma in a young girl presenting as a lytic lesion in the mandible. Surgical management with long term follow up is recommended to detect recurrence or malignant transformation. A careful differential diagnosis is warranted in cases of odontogenic tumours, especially in cases with unusual presentation.'

Keyword :
Ameloblastic fibroma, Odontogenic tumours, Odontoma, Hamartoma

Fig:1
Ameloblastic fibroma is a rare benign tumour which, according to WHO classification, belongs to the class of mixed odontogenic tumours. It is defined as a neoplasm with proliferating epithelium which is embedded in cellular ecto-mesenchymal tissue resembling dental papilla, and has varying degree of dental hard tissue formation & inductive changes [1]. Kruse first reported ameloblastic fibroma in 1891 [2]. Until recently, only about 123 cases of ameloblastic fibroma are reported in the literature [9]. We present a rare case of ameloblastic fibroma in a young girl presenting as a lytic lesion in the mandible.

**CASE REPORT:**
A 24 years old girl presented at our institute with a history of gradual increase in size of the swelling at left lower alveolar region over a period of 2 years. Examination revealed a 3x3cm sized sub mucosal swelling with mandibular irregularity in the region of left first molar tooth. Orthopentogram revealed radiolucent lesion in the left lower mandible. CT scan was suggestive of an expansile lytic lesion involving the lateral aspect of mandible on left side in the region of first molar tooth with partially impacted first molar tooth. No evidence of intracystic septations was seen. Patient already had a biopsy done elsewhere and the review of slides was suggestive of ameloblastic fibroma
In view of the lytic lesion, she underwent segmental resection of the left mandible and reconstruction using vascularized free fibular flap. Post-operative histopathology was suggestive of a 3x2.5x2.5 cm lesion in the left mandible with features suggestive of ameloblastic fibroma. Resected bone edges & marrow was free of tumour. Patient is on regular follow up & after two years of surgery, she is disease free and has excellent cosmetic outcome.

**DISCUSSION:** Ameloblastic fibroma is a rare benign tumour of odontogenic lineage with a relative frequency of 1.5% - 4.5% [6]. It most commonly occurs in first two decades of life [3], and is associated with tooth enclosure and a common site of occurrence is the posterior region of the mandible. Microscopic examination reveals strands, cords or islands of proliferating odontogenic epithelium admixed with primitive connective tissue stroma closely resembling the dental papilla. The clinical presentation of ameloblastic fibroma is a painless slow growing, expansile lesion of jaw which is accidentally discovered on radiographs in about 20% of cases. Typically, radiographs reveal a unilocular or multilocular radiolucent lesion, often with sclerotic borders [4,5]. Usually these lesions are slow growing causing expansion of the cortical plates rather that eroding them, but a peculiarity in our case was the presentation with lytic lesion in the mandible which is unusual for ameloblastic fibroma and thus malignant jaw tumour was also considered as a differential diagnosis. In addition to ameloblastic fibroma, the group of mixed odontogenic tumours also include related lesion like ameloblastic fibro-odontoma (AFO) and ameloblastic fibro-dentinoma (AFD)[6,7]. There is a debate regarding the exact nature of the ameloblastic fibroma & related lesions [6]. Cahn and Blum [8] proposed the continuum concept according to which ameloblastic fibroma can develop into an odontoma if allowed to remain and mature over time. Odontoma is a hamartomatous lesion that contains all calcified dental tissue. This concept thus implies that ameloblastic fibroma, AFO & AFD are different stages of the same lesion and will ultimately mature to form an odontoma. Some authors also believe that AFO is an intermediate stage in the formation of odontoma, the primary stage being ameloblastic fibroma [9]. However, there are oppositions on this continuum concept. First of all, ameloblastic fibroma occurs at an older age than AFO (mean age 9.6 years) which does not support the concept of ameloblastic fibroma developing into AFO & then to odontoma. Secondly, recurrent ameloblastic fibromas have not shown further differentiation into dental hard tissue forming odontoma. Lastly, ameloblastic fibroma occurs beyond the age of completion of odontogenesis, i.e, after 20 years. Ameloblastic fibroma is considered to be a true neoplasm with potential of recurrence and/or malignant transformation. However, a distinct type occurring in the childhood could be a primitive stage in the development of odontoma [10].As reported by Chen et al, 5 and 10 year rates of malignant transformation are 10.2% & 22.2% respectively [10]. A literature review by Muller et al [11] revealed that ameloblastic fibroma was the precursor lesion in 44% of ameloblastic fibrosarcomas developing since 1960. Also, the recurrence rates reported varies from 18.3% [12] to 43.5% [13]. The treatment for all cases of ameloblastic fibromas is exclusively surgical, with enucleation or curettage at one end and segmental resection with reconstruction at the other end for potentially malignant lesions as was

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done with an excellent cosmetic outcome in our patient. Regardless of the surgery performed, patients with ameloblastic fibroma should be closely followed up for long period of time in order to detect any recurrence or malignant transformation into ameloblastic fibrosarcoma.

CONCLUSION:
This report highlights the needs for a careful differential diagnosis in cases of odontogenic tumours, especially in cases with unusual presentation. Ameloblastic fibroma is a rare differential and clinicians should be aware of this entity so as to initiate a prompt and appropriate management.

REFERENCES:


