ODONTOAMELOBLASTOMA : A RARE ODONTOGENIC TUMOUR

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Introduction

The odontoameloblastoma is a rare mixed odontogenic tumour originating from proliferating epithelial and mesenchymal components characterized by simultaneous occurrence of ameloblastoma and composite odontoma [1-8]. So, few cases have been reported in literature till date that any statistical data regarding this disease may not be valid [6]. Approximately 45 cases have been reported in world literature but very few have the histologic and clinical criteria of odontoameloblastoma [1]. This lesion has various synonyms like ameloblastic odontoma, odontoblastoma, adamentto-odontoma and calcified mixed odontogenic tumour [6].

It usually occurs between 6 months to 40 years predominantly involving mandible with predilection for molar-premolar region [1,3]. It is an expansile centrally destructive lesion exhibiting slow growing characteristics like ameloblastoma and if left untreated may cause considerable facial deformity. Symptoms include a slowly progressive swelling of the alveolar plates, dull pain, an altered occlusion, delayed eruption or impacted teeth. Radiologically the tumor presents as a well defined unilocular or multilocular radiolucent cavity containing varying amounts of radio-opaque material, which may or may not bear resemblance to formed, albeit miniature teeth. It may also be in form of small, dense particles or as a large central mass leading to divergence of adjacent tooth roots [1,6].

Histologically the tumour consists of ameloblasts, enamel matrix, dentin, dental papilla, bone and cementum as well as stromal connective tissue. The classical picture is the presence of sheets of typical ameloblastoma of one or another type of recognized variety. The ameloblast cells may arrange themselves in cord or follicular pattern [6].

As this tumour resembles ameloblastoma in clinical behaviour aggressive treatment modality like en-bloc or segmental resection is advocated [6].

Case Report

36 year old patient was referred to maxillofacial clinic for evaluation and management of slowly growing swelling over right lower jaw since last 6 months. Examination revealed a bony hard, sessile, non-pulsatile, fixed, non-ulcerated, mildly tender diffuse swelling of 4x2 cm size extending from 46-41 region, involving both buccal and lingual cortex with 43 missing and 45 slightly mobile [Fig-1]. Mucosa over the swelling was mobile and normal in colour. Radiograph revealed a multilocular radiolucent lesion extending from 47-42 with multiple radio-opaque masses resembling tooth like structures. The histological examination revealed proliferating odontogenic epithelium in mature connective tissue stroma. The neoplastic odontogenic epithelium forming islands and cords between the dysplastic dentinoid and enamel with reverse nuclear polarization, typical of follicular ameloblastoma was observed (Fig-2). The patient was managed with en-bloc resection of the tumor mass along with 1 cm of healthy margin preserving the continuity of the inferior border of mandible. Cornoy's solution was applied to resected margin and frozen section was carried out to rule out any left over neoplastic tissue. The resected specimen was further examined to confirm the diagnosis. The primary reconstruction was not carried out as most of the recurrences have been reported to occur within first 3 months of surgical intervention as per available literature. After 3rd post-operative month the

Fig. 1 : Intra-oral view of the lesion

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Fig. 2 : Histopathological picture of odonto-ameloblastoma

patient was taken up successfully for mandibular reconstruction with autogenous cortico-cancellous iliac crest bone graft (Fig-3). The patient was evaluated periodically for any recurrence for one year.

Discussion

Odontoameloblastoma or ameloblastic odontoma has generated much controversy and confusion in literature as evidenced by reporting of ameloblastic fibro-odontomas, developing odontomas and odontoameloblastomas under the terms of ameloblastic odontoma. Choukus and Tots (1964) suggested that the ameloblastoma and odontoma may develop separately and due to invasive growth of the former odontoma becomes surrounded by the more aggressive ameloblastoma thus producing a true collision tumour. To clarify the confusion, WHO subdivided the category into ameloblastic fibro-odontoma and odontoameloblastoma based on their clinical course and histological picture [9]. Ameloblastic fibro-odontoma is a slow growing tumour, which can be treated effectively by enucleation. It does not spread between bony trabeculae and has low recurrence rate. However, odontoameloblastoma is a locally invasive, aggressive odontogenic tumour, which spreads by infiltrating between the bony trabeculae. This characteristic was also observed in our case both clinically and histopathologically.

From the histological picture of the lesion it is apparent that the hard tissue formed, are of hamartomatous nature due to presence of enamel, dentin and pulpal tissue. However, ameloblastoma is intermingled with the hard tissue forming a tumorous mass.

Fig. 3 : Mandibular reconstruction with autogenous iliac crest bone graft

The tumour behaves like ameloblastoma and tends to recur particularly when treated conservatively. The effective methods of treatment are en-bloc resection or complete resection of affected part of bone irrespective of size of the lesion. Early and periodic follow-up is also advised to detect any possible relapse.

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