CASE REPORT

Adolescent Mandibular Central Odontoameloblastoma: A Rare Case Report

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Abstract Odontoameloblastoma is a rare odontogenic tumour, characterised by simultaneous occurrence of an ameloblastoma and a compound or complex odontoma in the same tumoral mass. The tumour is seen in first three decades and affects mandible or maxilla equally, commonly found posterior to the canines. The management is similar to unicystic ameloblastoma and odontoma excision. A long term follow up is a must to observe the recurrence. Here we present a rare case of odontoameloblastoma in a 17 year old male, presenting as an asymptomatic anterior mandibular swelling with chief complaint of missing lower front teeth.

Keywords Odontoblastoma · Admantodontoma · Calcified mixed odontogenic tumour · Collision tumor · Ameloblasticodontoma

Introduction

Odontoameloblastoma (OA) is an extremely rare mixed odontogenic tumor often associated with an impacted

tooth, composed of neoplastic epithelium and myxomatous connective tissue [1, 2]. As these tumors resemble ameloblastoma in clinical behavior, both enucleation followed by chemical cauterization and aggressive treatment modality like en-bloc or segmental resection is advocated [3, 4, 5, 6].

Case Report

A 17 year old male patient reported for unerupted lower front teeth with asymptomatic swelling in lower chin. A diffuse 5 × 6 cm bony hard swelling in the chin region was present with no secondary skin changes (Fig. 1). Intra-orally labial and buccal cortex were expanded with normal color and texture of mucosa. Mandibular right central incisor, lateral incisor and canine were missing with mesial migration of left central incisor; all teeth were vital and non-mobile (Fig. 2). Imaging demonstrated an irregular shaped dense radiopaque mass, surrounded by a halo of radiolucency with definite boundary and impacted teeth adjacent to the tumour capsule (Fig. 3). An incisional biopsy reported follicular ameloblastoma and compound odontoma. A complete surgical enucleation was performed and all three impacted teeth were removed (Fig. 4a, b). The lateral clearance was adequate as we had to extract the impacted teeth on either side of the tumor margin. Lingual plate was thick enough to trim at least 1 mm of bone. Inferior margin was not possible due to inadequate bone on either side of midline approx. 2–3 cm out of 7 cm tumor dimension. The chemical cauterization with Cornoy's further destroys tumor cells to a depth of 1.5 mm from post enucleation bed. Eyelets were placed in upper and lower arch after completion of procedure. Patient was put on nasogastric tube feeds for 10 days. Maxilla-mandibular fixation was done on third post op day and continued for a

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Fig. 1 Preoperative diffuse swelling of chin

Fig. 2 Preoperative intraoral swelling of buccal/lingual cortical bone with mesially drifted left central and incisors

period of 6 weeks. Histological sections (10×) revealed numerous odontogenic epithelial islands and follicles within a fibro-cellular connective tissue. At 40×, the follicles showed peripheral tall columnar ameloblast like cells within central areas showing stellate reticulum like

Fig. 3 Preoperative orthopantomogram showing mixed tumor crossing the midline with irregular radiopaque central mass surrounded by halo radiolucency with impacted mandibular right central incisor, lateral incisor and canine

areas. The fibro-cellular connective tissue stroma also exhibited eosinophilic and basophilic calcifications in the form of multiple denticles (Fig. 5a–d). After 2 years of observation for recurrence, two implants were successfully placed with good osseointegration in spontaneously regenerated bone after orthodontic uprighting of adjacent teeth (Fig. 6).

Discussion

The term odontoameloblastoma was included in the year 1971 by World Health Organization (WHO) classification and is defined by WHO and Philipsen and Reichart as “a neoplasm that includes odontogenic ectomesenchyme in addition to odontogenic epithelium that resembles an ameloblastoma in both structure and behaviour [4]. It is a rare mixed odontogenic neoplasm, characterized by the simultaneous occurrence of an ameloblastoma and a compound or complex odontoma in the same tumour mass [7, 8]. It is being mentioned in literature by several names that include odontoblastoma, adamantodontoma, calcified mixed odontogenic tumour, soft and calcified odontoma, ameloblasticodontoma [4]. The final diagnosis for this particular tumour must be made based on the clinical radiological histopathological features [5].

The relative frequency of odontoameloblastoma among the odontogenic tumour varies between 1 and 3.1 % [9]. The pathogenesis of OA is unknown. One possible explanation is that the mineralized dental tissues are formed as a

Fig. 4 a, b Surgical exposure and excision of tumor

Fig. 5 a–d Histological sections ($\times 10$) reveal numerous odontogenic epithelial islands and follicles within a fibro-cellular connective tissue. On higher magnifications ($\times 40$), the follicles show peripheral tall columnar ameloblast like cells within central areas showing stellate reticulum like areas. The fibro-cellular connective tissue stroma also exhibit eosinophilic and basophilic calcifications in form of multiple denticles

hamartomatous proliferation in response to inductive stimuli produced by the proliferating epithelium over the mesenchymal tissue [2, 10, 11]. Odontoameloblastoma affects predominantly young patients with a median age of 20.12 years; in fact 59 % patients are under the age of 15 years [9]. Clinically OA starts as a slow growing painless mass that expands the alveolus and vestibular cortex, and prevents the eruption of the permanent teeth [8]. Radiologic examination of OA usually reveals a multilocular radiolucency with radiopaque areas resembling mature dental tissue [1, 5]. It might exhibit a well-defined margin, displacing the surrounding erupted teeth rather

than producing root resorption [8]. Some of the tumours may be relatively small when first detected (1–2 cm in diameter) [9]. Some of the OA may be exceedingly large, involving a considerable portion of the body of the mandible or maxilla [12]. Only three cases have been reported involving the anterior segment of the mandible. Review of the literature shows that, this tumour usually occurs in the posterior segments, with slight predilection for the mandible [4, 5].

Few OAs may contain ghost cells [13, 14]. It is important to exclude tumours that also contain such cells, namely, calcifying odontogenic cysts and odontogenic

Fig. 6 Two years postoperative orthopantomogram with implant placement in spontaneously regenerated bone for dental rehabilitation

ghost cell tumours [15]. Histologically, it is difficult to differentiate between odontoameloblastoma, ameloblastic fibro-odontoma and ameloblastic fibro-dentinoma [16]. Ameloblastic fibro-odontoma and ameloblastic fibro-dentinoma are slow-growing lesions with no propensity for bony expansion, mostly associated with an unerupted tooth [15]. Ameloblastic fibro-dentinoma and ameloblastic fibro-odontoma can be treated with enucleation without much danger of recurrence. Odontoameloblastoma on the other hand, expands by infiltrating between bony trabeculae, has a high rate of recurrence and should be aggressively treated like conventional ameloblastoma and close follow-up should be done [17].

A challenging subject with the treatment choice of OA cases is the decision whether to remove the teeth present in the affected areas of the tumour or not. Surgical enucleation is considered to be the treatment of choice in many cases with simultaneous removal of any involved unerupted tooth, since there are case reports of recurrence when conservative treatment with an attempt to save the associated teeth is performed [18–20]. The potential for odontoameloblastoma to recur is well-known. Yamamoto et al. [21] demonstrated a high proliferation potential of the odontoameloblastoma based on the expression of tenascin in the basement membrane of the odontogenic epithelium of this tumour and on the results obtained with proliferating cell nuclear antigen (PCNA) staining. These results indicate that this tumour may have the same biologic potential as that of an ameloblastoma and should therefore be treated and followed up in a similar fashion [2, 22]. The enucleation, ostectomy and chemical cauterization using Cornoy's was decided due to following reasons: 1. Young patient with a lesion in anterior mandible crossing midline extending from premolar to premolar measuring approx.

7 cm. 2. Reconstruction difficulties for anterior mandible defects as safe marginal resection would have left a defect of nearly 9 cm requiring microvascular flaps adding to the morbidity. 3. Patient's non acceptance for resection and reconstruction after discussion and opting for conservative management understanding the recurrence possibilities. 4. Given the rate of recurrence in 5 years, we took a chance of conservative management and watched carefully as patient was compliant with regular follow up.

Differential diagnosis must be made with mixed radiographic patterns such as ameloblastic fibro-odontoma, calcifying epithelial odontogenic tumour (CEOT), and odontoma. Ameloblastic fibro-odontoma affects adult patients in a slow growing fashion and exhibits a well defined capsule and a lack of infiltration of surrounding bone [15]. This permits an easy surgical enucleation with a low recurrence potential. Histologically, it presents immature dental tissue “papilla-like” with an absence of ameloblastic differentiation. The ameloblastic fibro-odontoma usually affects adult subjects with a slow-growing mass without bone infiltration. The CEOT is a benign but locally aggressive neoplasm. Its origin is actually uncertain as it may arise from reduced enamel epithelium or stratum intermedium of the tooth bud. Commonly occurs in the molar region of the mandible with lower frequency in premolar and anterior region. In about 50 % of cases the lesion is associated with an unerupted or impacted tooth. Histologically the CEOT presents islands of eosinophilic epithelial cells infiltrating the bony trabeculae. The cells present nuclear hyperchromatism and pleomorphism. Another typical aspect of CEOT is the presence of Liesegang's rings (concentric spherical calcification) [15].

Choukous 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 tumor [23]. The early ameloblastic changes within the cyst wall were first described by Vickers and Gorlin in 1970, and their histologic criteria for the diagnosis of unicystic ameloblastoma includes a cyst lined by ameloblastic epithelium with a tall columnar basal layer, subnuclear vacuoles, reverse polarity of hyperchromatic nucleus, and a thin layer of oedematous, degenerating stellate reticulum-like cells on the surface. As the lesion in our patient met all these criteria he diagnosed as a case of odonto-ameloblastoma. Treatment wise, OA should be treated by the wide excision and closely followed up for at least 5 years, as there is a high rate of recurrence [24, 25].

Conflict of interest None.

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