CASE REPORT

Ameloblastic fibro-odontoma

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SUMMARY

Ameloblastic fibro-odontoma is a slow growing, benign, expansile epithelial odontogenic tumour with odontogenic mesenchyme, accounting for 0.3–1.7% of jaw tumours, signifying its rarity. The WHO defines it as “a neoplasm composed of proliferating odontogenic epithelium in a cellular ectomesenchymal tissue with varying degrees of inductive changes and dental hard tissue formation”. We report a case of an 11-year-old girl who presented to the Department of Maxillo-Facial Medicine and Radiology for the evaluation of a swelling in the left posterior mandible. Her clinical chart and investigations unveiled it as ameloblastic fibro-odontoma. After a promising presurgical evaluation, the lesion was enucleated using an intraoral approach followed by osteoplasty. Osteogenesis was attained despite of any definitive techniques to promote bone regeneration. Immediate postoperative inter-maxillary fixation was performed to prevent pathological fractures for a period of 3 weeks. In an 8-month follow-up, no untoward complications were noticed.

BACKGROUND

Ameloblastic fibro-odontoma (AFO) is a rare benign, expansile, mixed odontogenic tumour constituting 0.3–1.7% of all odontogenic jaw tumours. AFO is more common in the first two decades of life, with no significant gender inclination. It usually presents as a painless, slow growing mass

Figure 2 Clinical presentation shows an ill-defined, diffuse swelling on the left posterior body of the mandible with bi-cortical expansion.

hindering tooth eruption. AFO has equal predilection for the mandible or maxilla and favours the posterior areas.¹

As stated in the literature review, tumour enucleation remains a wiser treatment of choice. In the present case of an 11-year-old girl, the tumour was enucleated through an intraoral approach followed by osteoplasty.² Follow-up investigations at 8-month follow-up revealed appreciable osteogenesis owing to the age of the patient and conservative surgical approach. Conventional enucleation without osteogenic prompting techniques is a promising therapeutic modality as it maintains the cosmetic integrity in younger individuals with facial deformity (owing to tumour).³

CASE PRESENTATION

An 11-year-old girl was brought to the Department of Maxillo-Facial Medicine and Radiology for the

Figure 1 Extraoral picture depicting an ill-defined oval-shaped swelling on the left posterior body of the mandible altering the normal anatomic contour of the lower border.

Figure 3 Periapical view with respect to 75 and 36 reports a well-defined homogenous radiopaque mass distal to 75.

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Figure 4 Orthopantomogram showing mixed dentition status with a radiopaque mass distal to 75 radiated by radiolucent and sclerotic borders. The displacement of inferior alveolar nerve and altered contour of the lower border of the mandible can be appreciated.

evaluation of a swelling on the left side of the face and left lower back teeth region. History revealed that the onset began 5 months earlier and was insidious, initially asymptomatic, and slow growing. Though the lesion began asymptotically, there was a history of intermittent sudden, throbbing pain lasting 10 min for the past 15 days, which drove the patient's family to seek specialist opinion. Extraoral examination revealed asymmetry due to a swelling on the left lower 1/3rd of the face ([figure 1](#)). Clinical presentation revealed an ill-defined, diffuse, roughly oval-shaped 2.5×3 cm swelling on the left posterior body of the mandible. Superiorly, it was 2.5 cm short of an imaginary line extending from the left corner of the mouth to the left lobule of the ear. Inferiorly it was 1 cm below the lower border of the mandible marking an alteration in the normal anatomical pattern of the lower border of the mandible. The surface of the skin over the swelling was normal. Palpation revealed a swelling with bony hard consistency that was mildly tender on palpation. Intraoral examination revealed an ill-defined diffuse oval-shaped swelling, approximately 3.5×2.5 cm, causing marked cortical expansion and extending anteriorly from the distal surface of 75 and posteriorly

to the left retromolar region. Superiorly, the swelling was at the level of the mandibular occlusal plane and inferiorly to the vestibule, causing obliteration ([figure 2](#)). Mucosal colour was normal to the adjacent mucosa and palpation revealed marked expansion of the buccolingual cortices, which were hard in consistency and mildly tender on palpation. Other notable findings were clinically missing 36 and paraesthesia. Medical, social and family history remained insignificant.

INVESTIGATIONS

Periapical view with respect to 75, 36 revealed a well-defined homogenous radiopaque mass distal to 75, inferior to where the crown of 36 could be noticed ([figure 3](#)). Orthopantomogram revealed mixed dentition with a radiopaque mass distal to 75, approximately 2.5×3.5 cms, radiated by radiolucent and sclerotic borders suggestive of a fibrous capsule inferior to which impacted 36 could be noticed. The displacement of inferior alveolar nerve and altered contour of the lower border of the mandible could be seen ([figure 4](#)). CT-axial section revealed an expansile lesion with a hyperdense area surrounded by hypodense rim in the left body of the mandible ([figure 5A](#)). Three-dimensional (3D) reconstructed imaging illustrated an altered lower border of the mandible with thin lingual cortex ([figure 5B](#)). 3D-reconstructed sagittal imaging revealed a hyperdense area surrounded by a hypodense rim (suggestive of fibrous-capsule) inferior to which the impacted tooth was seen, with varying radiodensities of the hyperdense mass and tooth proper ([figure 5C](#)). Microscopic evaluation of the enucleated section after H&E staining revealed highly cellular connective tissue with rounded or angular cells and a few delicate collagen fibres resembling dental papilla. The odontogenic epithelium was noted to be in the form of strands, cords and islands. The peripheral cells were tall and columnar, with hyperchromatic nuclei and reversed polarity resembling ameloblasts, and the central cells were loosely arranged, resembling stellate reticulum. The decalcified H&E stained section revealed an odontoma component with dentin, containing dentinal tubules, a scalloped

Figure 5 (A) CT-axial section revealing expanded cortices with a hyperdense area surrounded by hypodense rim in the left body of the mandible. (B) Three-dimensional (3D) reconstructed image illustrating altered lower border of the mandible with thin lingual cortex. (C): 3D-reconstructed sagittal view presenting hyperdense area surrounded by a hypodense rim (suggestive of fibrous-capsule) inferior to which is the impacted tooth with varying radiodensities of the hyperdense mass and tooth proper.

Figure 6 (A) Photomicrograph showing islands and strands of odontogenic epithelium lined by tall columnar cells resembling ameloblasts and central stellate reticulum-like tissue scattered within a highly cellular connective tissue resembling primitive dental papilla. (B) Decalcified H&E stained section revealing odontoma component with dentin containing dentinal tubules, scalloped dentinoenamel junction, enamel space and tissue resembling pulp.

dentinoenamel junction, enamel space and tissue resembling pulp (figure 6A, B).

DIFFERENTIAL DIAGNOSIS

Considering the clinical presentation and radiographic evaluation, a provisional diagnosis of odontoma was made. Odontoma was considered provisionally as it is the most common odontogenic tumour of the jaws, occurs during the early second decade, has female inclination favouring the posterior mandible (complex type) and is usually a reason for the failure of a tooth to erupt. Calcifying epithelial odontogenic tumour and AFO were considered under differential diagnosis.

- ▶ Calcifying epithelial odontogenic tumours (Pindborg tumours) have a definite predilection for the mandible and mostly develop in the premolar–molar area, with 52% association with unerupted or impacted teeth. Jaw expansion is the only symptom and palpation reveals a hard tumour. In the present case, though most of the clinical findings were inclined towards a calcifying epithelial odontogenic tumour, its occurrence in the early second decades of life is very unusual.
- ▶ AFOs are mixed odontogenic tumours of the jaws occurring in the second decades of life, with no gender predilection. They can occur equally in the maxilla and mandible, and favour the posterior areas; they are associated with unerupted/impacted teeth with epicentre lying occlusal to the developing teeth with perceptible cortical expansion. Despite the similarities, paraesthesia and radiological presentation as a conglomerate mass is relatively rare.^{4 5}

TREATMENT

After a promising presurgical evaluation, the procedure was carried out under general anaesthesia. Access was gained through an intraoral approach by placing crevicular incisions from 74 extending distally along the crest of the alveolar ridge ending in a distal-releasing incision along the crest of the

Figure 7 (A) Surgical session showing the approach gained intraorally. (B) Intraoperative photograph after enucleation of the lesion. (C) Enucleated tumour along with the impacted 36.

Figure 8 (A) Postoperative orthopantomogram. (B): Follow-up orthopantomogram after 8 months.

ascending ramus of the mandible (figure 7A). Enucleation of the lesion along with the impacted 36 was carried out followed by osteoplasty (figure 7B, C). No specific technique was employed to promote osteogenesis. Primary closure was performed with 3-0 vicryl suture material. To prevent pathological fracture post-surgery, the jaws were immobilised using ivy-islet intermaxillary fixation for a period of 3 weeks (figure 8A).

OUTCOME AND FOLLOW-UP

The patient has been under regular periodic review for the past 8 months, and is symptom-free. Even though no special treatment was given to initiate/promote bone formation, osteogenesis occurred, owing to the young age and precise technique (figure 8B and figure 9).

Figure 9 Follow-up intraoral picture after 8 months.

DISCUSSION

AFO is a benign, expansile, mixed odontogenic jaw tumour.⁵ A clear distinction has been drawn between AFO and ameloblastic odontoma, a lesion with similar nomenclature. Earlier, there was ambiguity about whether ameloblastic fibroma, AFO and complex odontoma were different entities or whether they represented various stages of the same lesion. Microscopically, the least differentiated tumour, ameloblastic fibroma, leads to AFO and eventually into complex odontoma. AFO represents an immature complex odontoma, and therefore represents a hamartoma rather than a neoplastic odontogenic lesion.^{1 6} AFO is relatively uncommon, with a prevalence of 1–3% among odontogenic tumours. The literature reveals that it occurs predominantly in children and young adults (usually in the first two-decades of life). It has an equal occurrence in upper and lower jaws and no gender specificity is noted. Despite its ability to inhibit tooth eruption or displace involved teeth, the teeth in the affected area remain vital.⁷ The clinical presentation of the tumour shows marked variations, ranging from lesions detectable only by histopathology, to large tumours with extensive calcified masses. A recent WHO classification of Odontogenic Tumours, published in 2005, refers to AFO as a benign lesion with no invasive course.⁸ Radiographically, it presents as a well-defined mixed radiological lesion or a conglomerate radiopaque mass. Nevertheless, confirmatory diagnosis is made according to microscopic study, demonstrating islands of odontogenic epithelium embedded in cell-rich ectomesenchyme similar to dental papilla.⁹ As mentioned in the literature review, enucleation still remains the golden standard in the treatment of AFO, except in cases of larger lesions, which are treated by extensive surgery. Preservation of the unerupted tooth or its removal depends on the clinical situation. Recurrence is rare and may happen owing to inadequate surgical removal, or if tumour remnants are retained in the resected margins, especially in cases with large tumours.^{2 3 5 10} In the present scenario, it was very difficult to differentiate the AFO from a developing complex odontoma, as they share a spectrum of common features with respect to their age of occurrence, site of presentation, lack of symptoms and their clinical association with an impacted tooth.

Learning points

- ▶ Considering ameloblastic fibro-odontoma in the differential diagnosis can guide us to a precise diagnosis in a clinical situation of a young patient with a swelling associated with an unerupted tooth favouring posterior regions.
- ▶ Conventional enucleation without osteogenic prompting techniques can be a promising therapeutic modality that retains the cosmetic integrity in younger individuals with facial deformity (owing to tumour).
- ▶ In the present case, a conservative surgical approach (enucleation) yielded superior results, further potentiating the literature review.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Gogri AA, Kadam SG, Umarji HR, *et al.* Ameloblastic fibro-odontoma differentiating into odontoma: an old concept revised. *J Indian Acad Oral Med Radiol* 2014;26:310–14.
- 2 De Riu G, Meloni SM, Contini M, *et al.* Ameloblastic fibro-odontoma. Case report and review of literature. *J Craniomaxillofac Surg* 2010;38:141–4.
- 3 Pontes HA, Pontes FS, Lameira AG, *et al.* Report of four cases of ameloblastic fibro-odontoma in mandible and discussion of the literature about the treatment. *J Craniomaxillofac Surg* 2012;40:e59–63.
- 4 Wood NK, Goaz PW, Kallal R. *Mixed radiolucent–radiopaque lesions associated with teeth in differential diagnosis of oral and maxillofacial lesions*. 5th edn. Elsevier. 2006:423–7.
- 5 Surej Kumar LK, Manuel S, Khalam SA, *et al.* Ameloblastic fibro-odontoma. *Int J Surg Case Rep* 2014;5:1142–4.
- 6 Rajendran R. *Cysts and tumors of odontogenic origin*. 6th edn. *Shafer's text book of oral pathology*, 2009:287.
- 7 deSouza Tolentino E, Centurion BS, Lima MC, *et al.* Ameloblastic fibro-odontoma: a diagnostic challenge. *Int J Dent* 2010;2010:pii: 104630.
- 8 Mummolo S, Marchetti E, Di Martinos S, *et al.* Ameloblastic fibro-odontoma: a case report. *Ann Stomatol (Roma)* 2010;1:11–13.
- 9 Dolanmaz D, Pampu AA, Kalayci A, *et al.* An unusual size of AmeloblasticFibro-odontoma. *DentomaxillofacRadiol* 2008;37:179–82.
- 10 Nayak B, Patnakar S, Khot K, *et al.* Ameloblastic fibro-odontoma of the maxilla: review of literature and report of a case. *J Indian Acad Oral Med Radiol* 2011;23:S442–4.

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