Rare disease

Large ameloblastic fibro-odontoma in an 18-year-old girl and review of literature

Ajay Pillai,¹ Swapnil Moghe,² Kanishka Navin Guru,³ Preeti P Nair⁴¹Department of Oral & Maxillofacial Surgery, People's Dental Academy, Bhopal, Madhya Pradesh, India²People's Dental Academy, Bhopal, Madhya Pradesh, India³Department of Oral Surgery, Peoples University, Bhopal, Madhya Pradesh, India⁴Department of Oral Medicine & Radiology, People's College of Dental Sciences & Research Centre, Bhopal, Madhya Pradesh, India

Correspondence to Preeti P Nair, shajihoss@gmail.com

Summary

The ameloblastic fibro-odontoma is a rare, benign, mixed neoplasm composed of proliferating odontogenic epithelium, ectomesenchymal tissue and varying degrees of dental hard tissue formation. It occurs exclusively as an intraosseous lesion. It usually exhibits slow growth and is commonly seen in children and young adults. Radiologically, ameloblastic fibro-odontoma appears as a circumscribed radiolucency which may contain radiopaque foci. Most cases of ameloblastic fibro-odontoma exhibit benign behaviour, but cases of malignant transformation have been reported. The treatment modality in most cases involves conservative surgery, but in cases with malignant transformation more radical treatment will be required. A massive ameloblastic fibro-odontoma involving the mandible is being described here with its clinical, radiological and histopathological features.

BACKGROUND

Ameloblastic fibro-odontoma (AFO) is a rare odontogenic tumour defined by WHO as a benign neoplasm composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles dental papilla, and with varying degrees of inductive change and dental hard tissue formation.¹ It is classified as tumours of odontogenic epithelium with odontogenic ectomesenchymal, with or without dental hard tissue formation.¹ The mean age of incidence is from 8 to 12 years and there is no gender predilection. The tumour is usually found in the mandible of children or teenagers. The relative arrangement of the soft tissues and the stage of development of the involved tooth are useful criteria for differential diagnosis. According to the revised WHO classification, it is a benign tumour without invasive growth, contrastingly to ameloblastoma.¹ The term AFO was first used by Hooker.² It is necessary to differentiate this lesion from odontoameloblastoma (OA).³ This distinction is important because of the occasional reports of the aggressive and recurrent nature of OA. Howell *et al*⁴ have shown the potential for malignant transformation of AFO to ameloblastic fibrosarcoma.

CASE PRESENTATION

A 18-year-old female patient was referred to the department of oral and maxillofacial surgery with a complaint a diffuse swelling in mandible. Clinical examination revealed a large swelling in left side of mandible resulting in facial asymmetry (figure 1).

Her medical, family and social history was unremarkable. There was no history of trauma. Oral examination revealed an irregular cauliflower-like outgrowth, stony hard in consistency, located in the retromolar area of left side of the mandible (figure 2). Patient revealed a history

of dental extraction of first molar of same side 6 months earlier. Second molar was clinically missing. There was no history of paraesthesia or anaesthesia. Panoramic radiography revealed a well-defined radiopaque lesion with radiolucent border and impacted tooth occupying a large

Figure 1 Facial profile.

Figure 2 Intra oral profile.

area (approximately 30×40 mm) of the ramus of the mandible (figure 3). It was clinically diagnosed as an odontoma.

INVESTIGATIONS

- ▶ Orthopantomogram (figure 3).
- ▶ Complete blood count.
- ▶ Incision biopsy performed. Histopathological examination (HPE) report suggestive of AFO.
- ▶ Excisional biopsy confirmed the report of the incisional biopsy.

Figure 3 Panoramic radiography showed lesion appeared as well-defined radiopaque with radiolucent area and impacted tooth within large area of ramus of the mandible.

DIFFERENTIAL DIAGNOSIS

Differential diagnosis included odontoma, ameloblastoma, OA and ameloblastic fibroma.

On the one hand, the presence of both dentin and enamel is important to differentiate the lesion from ameloblastic fibroma, which shows no sign of tooth differentiation.

On the other hand, the exclusive presence of dentin characterises another entity, ameloblastic fibrodentinoma.⁵

OA is usually an unencapsulated lesion, whose behaviour resembles that of ameloblastoma. Presence of ameloblastic areas and ghost cells aids in the differential diagnosis.¹

TREATMENT

The lesion was enucleated in toto along with an intact capsule under general anaesthesia (figure 4) and the specimen was sent for histopathological analysis.

Histopathology revealed an odontogenic neoplasia characterised by an odontogenic epithelium proliferation, island and strand-like that of varying sizes, with a peripheral rim of columnar cells resembling enamel epithelium, and a loosely arranged spindle-shaped epithelium identical to stellate reticulum in the central area. The lesion stroma showed stellate-shaped fibroblasts in a myxoid arrangement resembling dental papilla. Dentine-like material and enamel matrix were observed in other areas, which sometimes surrounded the odontogenic epithelial islands. The large amount of calcifying tissue presence was unusual considering the patient's age. No finding suggestive of malignant growth was observed. On the basis of the above findings, a histopathological diagnosis of AFO was arrived at (figure 5), which was consistent with that of the incisional biopsy.

An immunohistochemical study was not possible in our setup.

OUTCOME AND FOLLOW-UP

After 4-month follow-up period, no sign of recurrence was observed. The patient continues to be on regular follow-up.

DISCUSSION

Philipsen *et al*⁶ reported a series of 86 cases of AFO, in which only one case was reported above 20 years of age,

Figure 4 The lesion was enucleated.

Figure 5 Histopathological features suggestive of ameloblastic fibro-odontoma.

with an average age of 9 years (ranging from 1 to 22 years old), showing a male–female ratio of 1.4:1.

The most common site was the posterior mandible, with a mandible–maxilla ratio of 2:4. In this case report, the subject was an 18-year-old girl, the lesion was located in the posterior mandible. Usually, the lesion is incidentally detected when investigating cause for failed eruption of permanent dentition.⁶ The patient in this case report presented with expansion of the bone, but was otherwise asymptomatic.

The histological characteristics of AFO cannot be distinguished from odontoma, and consequently, some authors⁶ believe that AFO is an immature complex odontoma. In the same way, no ultra structural differences were observed between AFO and ameloblastic fibroma.⁶ The large amount of calcifying tissue present in the lesion may speak in favour of the theory that AFO is an early stage of odontoma. The amount of calcified material within the lesion is considerably variable. In the case presented, the formation of many denticles, similar to an odontoma was observed. The incidence of AFO varies from 0.3% to 1.7%^{7–8} among the odontogenic tumours, and when only paediatric cases are considered it represents 4.6%.⁸ The lesion presented during teenage, with formation of a large amount of calcifying tissue.

Radiographically, AFO shows radiopaque and radiolucent areas that vary from one lesion to another; sometimes large amounts of mineralised tissue may resemble an odontoma.⁸ Lesions can vary from a mostly radiolucent lesion to one that is nearly completely radiopaque. The radiographic features and localisation of the lesion in this case report, was suggestive of an odontoma.

Usually, AFO is well encapsulated and different from ameloblastoma, presenting no local invasion. This was the reason for a more conservative treatment, as recommended by literature.⁶ Since the lesion was well-encapsulated it was easy to create cleavage between bone and AFO. The lesion was removed without capsule laceration, which was better for prognosis. Enucleation of the lesion was a satisfactory with good prognosis. The results of the cell kinetics of AFO by bromhexidine and PCNA by Sekine *et al* suggests that the ectomesenchymal component was more proliferative than the epithelial

component.⁹ Immunohistochemical (IHC) studies are very informative,¹⁰ and useful in the diagnosis of the lesion as this lesion occasionally transforms into an aggressive type. However, we could not do an IHC study as it was not possible in our setup and the patient could not afford it in other laboratories. The recommended treatment for AFO is conservative surgery with enucleation. The potential for malignant transformation does not justify radical treatment.¹¹ If there is a recurrence accompanied by change of the histological pattern towards a more unorganised fibrous stroma, with displacement of the epithelial component, then more extensive treatment procedures appear to be indicated.¹² Though rare, malignant transformation may occur⁴ and long-term follow-up is recommended.¹³ A follow-up of only few months or years is generally inadequate to state that a definite cure has been achieved, as malignant changes can occur after 10–12 years also.¹⁴

Learning points

- ▶ Immunohistochemical study is very helpful to know the aggressive nature of tumour and its potential to turn malignant.
- ▶ As ameloblastic fibro-odontoma is an encapsulated tumour therefore a good surgical cleavage between tumour and bone is available and it is possible to remove the capsule without perforation.
- ▶ Ameloblastic fibro-odontoma show typical histopathological features of epithelial and ectomesenchymal origin with hard tissue deposition.
- ▶ The differential diagnosis with odontoameloblastoma and odontoma is essential for the correct treatment and follow-up, as well as, to reduce patient morbidity.
- ▶ It is necessary to differentiate this lesion from odontoameloblastoma. This distinction is important because of the occasional reports of the aggressive and recurrent nature of odontoameloblastoma.

Competing interests None.

Patient consent Obtained.

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