CASE REPORT

Mixed odontogenic tumour with dentinoid and ghost cells

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SUMMARY

Ameloblastomas do not generally show evidence of induction, however, rare cases associated with odontome have been reported and are referred to as odontoameloblastomas. We report an unusual case of an ameloblastoma with features of an adenomatoid odontogenic tumour, showing evidence of induction of dentinoid by tumour cells—but without concomitant formation of enamel—and with features of ghost cells. The lesion occurred on the left side of the maxilla in a 31-year-old woman.

BACKGROUND

Odontogenic cysts and tumours comprise a group of lesions that is rather diverse, undoubtedly because of their complex histogenetic source (histogenesis).¹ Odontogenic tumours composed of two distinct types of lesions are rare. As neoplastic and hamartomatous aberrations can occur at any stage of odontogenesis, combined features of odontogenic tumours may arise. Moreover, tumours that are classified into the group of epithelial tumours without odontogenic ectomesenchyme, such as ameloblastoma and adenomatoid odontogenic tumour (AOT), show evidence of dentinoid induction. Ameloblastoma is the most primitive odontogenic tumour and although it is characterised by a wide variety of histological appearances, this tumour does not generally show evidence of induction. With the exception of odontoameloblastoma, there is no dental hard tissue formation in ameloblastomas. However, rare cases of ameloblastomas showing evidence of dentinoid induction by tumour cells, but without concomitant formation of enamel, have been reported.² AOT is a benign, non-neoplastic (hamartomatous) lesion with a slow but progressive growth. It accounts for 3–7% of all odontogenic tumours and was once considered a histological variant of solid/multicystic ameloblastoma. Histopathologically, AOT is characterised by multisized solid nodules of odontogenic epithelial cells forming nests or rosette-like structures, and within the cellular areas are structures of tubular or duct-like appearance, which led to the term ‘adenameloblastoma’, previously used to designate this tumour.^{3,4} Some cases having combined histopathological features of ameloblastoma and AOT have been reported in the literature. In this article, we report a rare odontogenic tumour with similar histopathological features. This uncommon composite odontogenic tumour was first described by Brannon and suggested the name ‘Adenoid ameloblastoma with dentinoid’, as cited by

Ghasemi-Moridani and Yazdi,⁴ Evans *et al*⁵ and Matsumoto *et al*.⁶

The hybrid/mixed odontogenic tumour is a separate entity that does not come under any subtype of odontogenic tumour classification (WHO). These tumours are composed of more than one pathognomonic feature, which causes difficulty for the pathologist in delineating one tumour from the other. Slabbert *et al* first claimed a case of dentoameloblastoma with features of ameloblastoma and large amounts of a dentinoid-type of dystrophic calcification.² We present a similar case of hybrid/mixed odontogenic tumour comprising of features of both AOT and ameloblastoma.

CASE PRESENTATION

A 31-year-old woman visited the outpatient department of oral medicine and radiology with swelling on the left side of the face for 6 months. The patient gave a history of a peanut-sized swelling initially, which gradually increased to its present size. There was no history of pain or discharge. Her dental history revealed that the patient had a similar swelling 3 years prior in the same region, which was diagnosed as AOT, for which she had undergone surgical treatment in a private hospital; now, 3 years later, the swelling reappeared at the same site. Extra-oral findings included diffuse swelling on the left side of the face extending superiorly from the lower palpebrae, inferiorly to the lower border of the mandible and anteriorly from the midline of the face to 3 cm posteriorly out from the tragus of the ear, with slight obliteration of the nose, measuring about 6×4 cm in size and oval in shape (figure 1). Deviation of the nose to the right was evident. Eye opening was reduced slightly on the involved side. On palpation, swelling was hard in consistency and non-tender. Intraoral examination revealed diffuse swelling in the upper left alveolar region extending from the tooth region 21 to 26 (Federation Dentaire Internationale notation of tooth numbering) of the jaw (figure 2). Absence of teeth was noted in the left quadrant of 23–28. Alveolar expansion was present in a few areas, giving an impression of the presence of one single large swelling. Mild tenderness was evident on palpating the lesion. There was no redness on the surface of the skin. The overlying mucosa was continuous, showing no distinction of skin between mucosa covering the swelling and normal tissue.

INVESTIGATIONS

Radiographs (intraoral and extraoral images) were advised in the form of intraoral periapical view,

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Figure 1 Presence of large swelling in middle third of face with apparent deviation of nose.

orthopantomography, occlusal view (maxillary occlusal view) and CT.

The intraoral and extraoral radiography showed greater areas of radiolucency with complete lack of bone in the area of swelling (figures 3 and 4).

Axial and coronal CT showed a well-defined hyperdense area crossing the midline of the arch up to 12 (figure 5). Considering the clinical and radiological diagnosis, surgical treatment was performed under local anaesthesia.

TREATMENT AND FOLLOW-UP

Excisional biopsy was planned for allowing complete excision of the lesion. Later, the specimen was sent for histopathological diagnosis. The gross specimen measured 7 cm×5 cm×5 cm and

Figure 2 Intraoral image showing presence of large swelling in the maxillary left side.

Figure 3 Orthopantomographic view.

was irregular in shape; the associated teeth were 11, 12 and 21 (figure 6).

H&E section showed features suggestive of a collision between two odontogenic tumours, namely AOT and plexiform ameloblastoma. A few areas of the section showed plump spindle-shaped cells arranged as rosettes and ducts. The lining of the ducts showed tall columnar cells along with nucleus facing away from the luminal wall (figure 7). Some sections of the specimen showed odontogenic epithelial cells arranged in the form of an island. Inductive changes were seen adjacent to the tall columnar ameloblast-like cells, with reversal of polarity, and were arranged in a plexiform pattern resembling a plexiform ameloblastoma (figure 8). The stroma was loose with sheets of stellate reticulum-like cells also evident, with dentinoid-like deposition along with ghost cells (figure 9).

The H&E section had two distinct characteristics. The core of the gross specimen showed features of strands of odontogenic epithelial cells with tall columnar cells showing reversal of polarity of the nuclei. The central part of the strands showed stellate reticulum-like features. The loose connective tissue stroma showed dentinoid-like deposition along with ghost cells. As the whole of the specimen was kept in to processing, the other part of the specimen except the central core were showing plump spindle-shaped cells arranged in the form of rosettes and ducts. The lining cells of ducts showed tall columnar cells along with nucleus facing away from the luminal wall. Correlating the features of the entire gross specimen a diagnosis of adenoidameloblastoma (hybrid/mixed odontogenic tumour) was given.

Figure 4 Maxillary occlusal view.

Figure 5 CT image of involved area.

OUTCOME AND FOLLOW-UP

The patient was examined postsurgically and showed resolution of symptoms. She later underwent rehabilitation treatment for the lost tooth to restore normal oral function (figure 10).

Confirmatory diagnosis

The features were suggestive of mixed odontogenic tumour.

DISCUSSION

Dentinoameloblastoma was initially described by Slabbert *et al* when they detected a close relationship between a dentinoid-like material and odontogenic epithelium in unicystic neoplasms. These neoplasms contained characteristic follicles of ameloblastoma along with a psammomatous type of dystrophic calcification and were reported in an Asian male patient.² However, the present case showed plexiform ameloblastoma alongside a large amount of dentinoid/osteodentin-like material, and the additional and unique presence of ghost cells. Ghasemi-Moridani and Yazdi,⁴ and Sonone *et al*,⁷ reported similar cases of plexiform ameloblastoma and AOT along with hard tissue formation. Presence of ghost cells in ameloblastoma is not a common

Figure 7 Histological view of lesion.

feature, but ameloblastic fibrodontoma/odontomas have shown convincing evidence of the presence of ghost cells. Ghost cells that are present are enucleated cells, but they still keep the outline of the cell membrane intact. These cells undergo dystrophic calcification/mineralisation distinguished by fine basophilic granularity and, eventually, result in large sheets of calcified material. On occasion, ghost cells may get displaced in the connective tissue wall, drawing out a foreign-body giant cell response.⁸ Recently, Sonone *et al*⁷ also reported a case of adenoid ameloblastoma with dentinoid induction with presence of ghost cells. There are only 14 reported cases of odontogenic tumour that have histopathological features similar to ameloblastoma or AOT, or both, with the formation of hardened tissue resembling dentinoid. On reviewing the case reports, we observed a wide range in distribution of age ranging from 4 years 11 months to 42 years, with a mean age of 26.5 years. Of the 14 cases, 8 were male and 6 female. The most common site for occurrence was the posterior mandible. Tajima *et al* reported a case involving the maxillary antrum, whereas Ghasemi-Moridani and Yazdi reported a case of involvement in the right posterior side of the maxilla. In our case report, the patient was a 31-year-old woman^{4 9}—an age over that of the mean age of occurrence. In our patient, the lesion was in the posterior of the maxilla, which is an uncommon spot for occurrence of the lesion. From the histopathological features, the tentative diagnosis of adenomatoid ameloblastoma with dentinoid was made. The histopathological features of this composite

Figure 6 Gross specimen of excised mass.

Figure 8 High resolution view of lesion.

Figure 9 Dentinoid and ghost cells.

odontogenic tumour varied in various cases reported in the literature, with regard to hard tissue formation. The cases reported by Ghasemi-Moridani and Yazdi,⁴ Evans *et al*,⁵ Matsumoto *et al*,⁶ Sonone *et al*,⁷ Orlowski *et al*,¹⁰ Tajima *et al*,¹¹ Takeda,¹¹ Dunlap and Fritzlen,¹² and Kiran *et al* show histopathological features of AOT and ameloblastoma, most commonly plexiform. The cases reported by Allen *et al* and Slabbert *et al*² show features only of ameloblastoma. The cases reported by all the other authors showed the induction phenomenon or hard tissue formation, mostly of dentin with or without dentinal tubules or dentinoid-like material.¹ The nature of the hard tissue, whether dentinoid, enamel or both, presenting in association with odontogenic epithelium, is varied. Takeda reported formation of bone, whereas Dunlap and Fritzlen,¹² and Tajima *et al*,⁹ described the formation of enamel matrix; others have reported the formation of dentin or dentinoid.¹¹ This suggests the possibility that neoplastic cells of pure epithelial tumours are involved in the synthesis of hard tissue. There were no signs

Figure 10 Postoperative view.

of recurrence 6 months postoperatively; denture prosthesis was given to the patient. Follow-up in most of the reported cases showed no recurrence to a maximum of 8 years. Evans *et al*⁵ described a recurrent lesion that had initially been diagnosed as AOT. Later, after the third recurrence and reviewing of all histologic findings, they named it adenoid ameloblastoma with dentinoid.

CONCLUSION

Odontogenic cysts and tumours comprise a group of lesions that are very diverse. A thorough knowledge of these lesions is required, to differentiate one from the other, to provide a proper cure to patients.

Learning points

- ▶ Clinical and radiographic evaluation only produces a tentative diagnosis; histopathological examination should be carried out for final diagnosis.
- ▶ Hamartomatous growth can give rise to several tissues based on histogenesis and stage of histogenesis.
- ▶ Rehabilitation of the patient is of utmost importance apart from excising the lesion.

Competing interests None declared.

Patient consent Obtained.

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