The value of three-dimensional imaging in the detection of a case of double pathology in the right maxilla of a patient

SUMMARY

A 14 year-old Black female patient was referred for management of a lesion in her right maxilla. The lesion measured about 3cm in diameter and was localised lateral to the right ala of the nose. It felt bony-hard in some areas and soft in others, and was continuous with the maxillary buccal plate. The overlying skin was normal in colour and texture. There was no loss of sensation in the right cheek and the patient presented with neither lymphadenitis nor lymphadenopathy. The overlying skin was normal in colour and texture. There was no loss of sensation in the right cheek and the patient presented with neither lymphadenitis nor lymphadenopathy. The overlying skin was normal in colour and texture.
A panoramic radiograph showed a radiopaque mass surrounded by a radiolucent periphery extending from the 11 to about the 14 region and which had caused lateral displacement of the root of the 53. (Figure 1). The borders of the lesion were fairly well defined for most of its extent but in general there was a lack of clarity. An impacted and ectopic 13 was present, lying between the roots of 15 and 16. Radiopaque lesions in the third quadrant were also detected but were diagnosed as dense bone islands and considered to be of no significance.

A computed tomographic scan was requested which revealed two separate lesions, one lying buccal and the other lying palatal. The buccal lesion was seen to be a well-defined unilocular radiolucency containing a radiopaque mass (Figure 2).

Palatal to this lesion but slightly more distal was an impacted 13 with a small cystic lesion that appeared to be attached to its cervical margin (Figure 3A) and which had caused thinning and slight expansion of the palatal plate. The two lesions were separated by a thin bony margin. (Figure 3B).

Both lesions were deemed to be of odontogenic origin as they were situated outside the maxillary sinus. For the buccal lesion, a differential diagnosis of calcifying odontogenic cyst, adenomatoid odontogenic tumour and ameloblastic fibro-odontoma was considered, all of these lesions being associated with radiopaque masses.

The palatal lesion associated with the impacted tooth was provisionally diagnosed as a dentigerous (follicular) cyst or, far less likely, an odontogenic keratocyst. An adenomatoid odontogenic tumour in the early stage before the development of dental hard tissue was considered, to avoid the possibility of double pathology.

A full-thickness mucoperiosteal midline flap was raised and the lesion easily separated from the flap. In some areas the bone over the lesion was paper thin. The buccal lesion appeared cystic, and was enucleated together with its bony hard contents. The impacted 13 could then be visualised through very thin bone. The 13 was sectioned and the crown removed together with the attached cyst. The root of the 13 was unfortunately dislodged into the maxillary sinus while the crown was elevated, but was easily recovered. Peripheral ostectomy was performed with copious irrigation and the wound closed with 3.0 vicryl suture.

Histological examination confirmed the buccal lesion to be a calcifying odontogenic cyst and the palatal lesion was shown to be a dentigerous cyst. The patient had an uneventful recovery.

HISTORICAL REVIEW

The calcifying odontogenic cyst has evoked much interest and controversy over the past five decades. It was described by Gorlin and his co-workers in 1962 as a developmental cyst which may exhibit aggressive behaviour, particularly the solid variety. The lesion appeared to exhibit features of an odontogenic tumour and was later regarded as such by the World Health Organisation. Praetorius and co-workers in 1981 classified the lesion into cystic and solid varieties. They designated the cystic variety as Type 1 and the solid variety as Type 2. Further, they named the solid variety a dentinogenic ghost cell tumour as, apart from the presence of epithelial ghost cells, they noticed what appeared to be dentinoid adjacent to the epithelium. Since then the solid type has evoked a variety of different names viz. calcifying ghost cell odontogenic cyst, ghost cell odontogenic tumour and calcifying cystic odontogenic tumour. Since the solid intrabony variety is less common than the cystic type and the lesions display a variable range of clinical behaviours, it is still widely recognised by many as a cyst.
Histologically the lesion presents with a fibrous capsule and a lining of odontogenic epithelium within which are found ghost cells. The latter are altered epithelial cells which have lost their nuclei and are a characteristic feature of the lesion. Calcification occurs within the ghost cells. In the solid form of the lesion the lumen is usually filled with islands of odontogenic epithelium within a fibrous stroma containing numerous ghost cells. The epithelial islands often resemble those found in an ameloblastoma. Dentinoid adjacent to the epithelium has been reported by some authors.

The dentigerous cyst on the other hand, by definition, surrounds the crown of an unerupted tooth, being attached to its neck, and has an epithelial lining derived from the dental follicle. As such it is merely a reduced enamel epithelium with no potential to proliferate. For this reason the cyst is non-aggressive and will not recur if any cells are left behind following enucleation. Under certain circumstances marsupialisation may be the treatment of choice.

This case of dual pathology illustrates the benefits and value of three-dimensional imaging in the diagnosis and management of these rare presentations.

Conflict of Interest: None declared

References