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

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SUMMARY

A 14 year-old Black female was referred for management of an asymptomatic swelling in her right maxilla. The lesion measured 3cm across, was localised lateral to the right ala of the nose, felt bony hard in some areas and soft in others, and was continuous with the maxillary buccal plate. It occupied the right anterior vestibule but there was no palatal expansion. A panoramic radiograph showed a radiopaque lesion surrounded by a radiolucent periphery, but a lack of clarity prompted a computed tomographic scan. The latter revealed two separate lesions, one buccal and one palatal. The buccal lesion showed a well-defined radiolucency containing a radiopaque mass while the palatal lesion showed a small cystic area attached to the neck of an impacted tooth. Differential diagnoses of calcifying odontogenic cyst, adenomatoid odontogenic tumour or ameloblastic fibro-odontoma and dentigerous cyst or odontogenic keratocyst were considered for the two lesions respectively. Enucleation of the buccal lesion and removal of the impacted tooth together with the overlying cyst presented no problem. Histologically the lesions were respectively diagnosed as a calcifying odontogenic cyst and a dentigerous cyst. Histological features are briefly described together with an historical review of the calcifying odontogenic cyst which has evoked much interest and controversy over the past five decades.

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INTRODUCTION

A tooth germ during its entire developmental period may be subject to many divergences from normal, resulting in a large spectrum of malformations. Some of these may include abnormal proliferations of the odontogenic epithelium producing cellular tumours (eg ameloblastoma) or tumours containing masses of calcified dental tissues. Alternatively, remnants of the odontogenic epithelium may produce cysts.[1] Despite the many possibilities of divergence from normal, the course of nature nevertheless "runs according to plan" since abnormalities are relatively rare as compared with the number of individuals with normal dentitions free from cysts or tumours.

One may logically reason that the chances of tooth buds diverging from normal in two different directions resulting in dual pathology would be rarer still. This fact is borne out in the paucity of reported cases of dual pathology in a maxillofacial setting as seen in the dental literature.

The following case report, in addition to describing a rare occurrence of this nature, highlights the importance of three-dimensional imaging in identifying the true nature of the lesion, which displayed lack of clarity on a panoramic radiograph.

CASE REPORT

A 14 year-old Black female patient was referred for management of a lesion in her right maxilla. The patient complained of a slight swelling on the right side of her face which she had first noticed about three months previously. It was asymptomatic. 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 not fixed to the overlying soft tissues, being rather continuous with the maxillary buccal plate of bone. 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. Intraorally the mucosa was normal in colour with no ulceration. The lesion occupied the right anterior vestibule but there was no palatal expansion. Teeth 12 and 13 were absent but retained 52 and 53 were present.

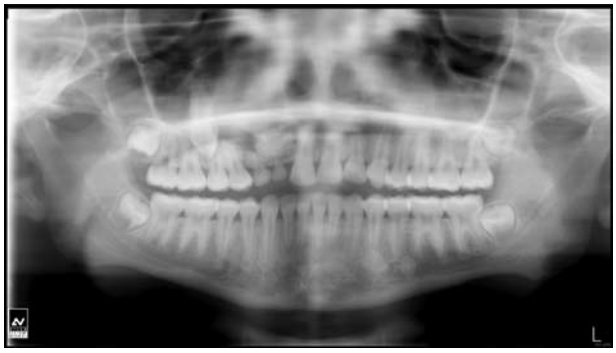

Figure 1: Panoramic radiograph illustrating the lesion

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.[2] The lesion appeared to exhibit features of an odontogenic tumour and was later regarded as such by the World Health Organisation.[3] 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.[4] 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.[5,6] 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.[6]

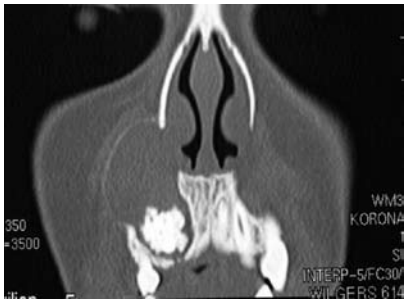

Figure 2: A computed tomographic coronal section scan illustrating the lesion

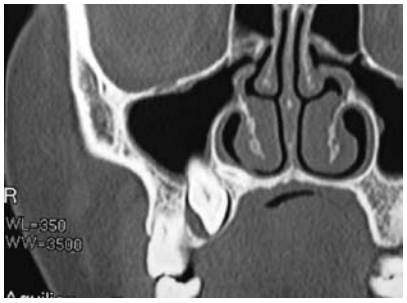

Figure 3A: Axial CT scan showing an impacted 13 with pericoronal lesion

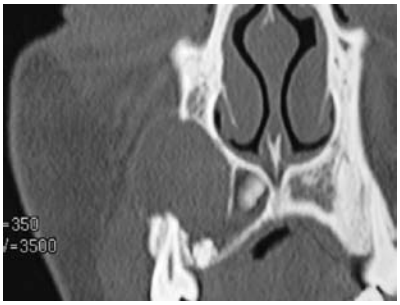

Figure 3B: Axial CT scan showing both buccal and palatal lesions

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.[6]

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.[7]

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

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