CASE REPORT

Unicystic ameloblastoma arising from a residual cyst

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SUMMARY

Intraoral swellings involving alveolar ridges in edentulous patients are clinically diagnosed as residual cysts, traumatic bone cysts, Stafne's jaw bone cavity, ameloblastoma and metastatic tumours of the jaw. This case report describes a residual cyst in a 68-year-old edentulous male patient which was enucleated and histopathologically confirmed as a unicystic ameloblastoma.

BACKGROUND

Periapical pathologies of the teeth are generally neglected by dental practitioners. Carious teeth with periapical pathologies are treated by either extraction or root canal treatment. After the completion of either, they do not follow-up the patient presuming that the treatment has been successful in resolving the pathology, as the patient is asymptomatic. This negligence can be fatal to patients as the pathology might not undergo spontaneous resolution and may transform into a residual cyst that may later give rise to a tumour such as ameloblastoma, adenomatoid odontogenic tumour or squamous cell carcinoma.¹⁻³ Hence, this case highlights the importance of postoperative follow-up of patients and the need for histopathological investigation to prevent recurrences of potential neoplastic transformations.

CASE PRESENTATION

A 68-year-old male patient was referred to the oral and maxillofacial surgeon with swelling in the right lower back region of the jaw since the past 6 months. The patient was relatively asymptomatic before 6 months when he noticed a swelling in the lower right back region of the jaw which was initially small in size and rapidly enlarged to the present size. No pus, discharge or pain was present. The patient had a history of extraction of all teeth 2 years previously due to caries and periodontal involvement. There was no relevant medical history reported.

The extraoral examination revealed a bilaterally symmetrical face, with no relevant findings. On intraoral examination, the patient was found to be completely edentulous. A round/oval-shaped swelling measuring about 2 cm×2 cm was present in the right lower back region on the alveolar ridge obliterating the buccal and lingual vestibules in the premolar area (figure 1). No signs of pus discharge or sinus tract were present. The surface of the swelling was smooth and the borders were well defined and regular without any ulceration. The swelling was soft to firm and non-tender (figure 1).

Figure 1 Intraoral photograph of the patient showing the lesion on the edentulous ridge.

INVESTIGATIONS

A CT was performed that showed an oval radiolucency surrounded by a well-defined radiopaque margin in the lower part. Resorption of the buccal and lingual portions of the edentulous alveolar ridge was present. Expansion on the buccal side was more than on the lingual side (figure 2).

DIFFERENTIAL DIAGNOSIS

- ▶ Residual cyst
- ▶ Aneurysmal bone cyst
- ▶ Metastatic tumour to the jaws
- ▶ Traumatic bone cyst
- ▶ Ameloblastoma
- ▶ Keratocystic odontogenic tumour

TREATMENT

The patient was operated under local anaesthesia using an inferior alveolar nerve block. A crestal incision was made on the swelling in the midline starting anteriorly from the symphysis region extending posteriorly 1 cm distal to the swelling. The mucosa was dissected and the cystic wall was visualised. The entire cyst was enucleated in toto. Excess soft tissue was cut and closure was carried out with a 3-0 silk suture. The specimen was sent for histopathological evaluation.

On gross examination, the tissue specimen was found to measure approximately 1.5×3.5 cm and be roughly oval in shape and whitish brown in colour. It was firm in consistency (figure 3) and was cut into two halves. The cystic lumen containing a whitish proliferative mass could be seen.

On microscopic examination, the H&E-stained section showed a cystic cavity lined by a non-keratinised stratified squamous epithelium. The epithelium had a characteristic arched pattern and, at focal areas, luminal proliferation was noted. On

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Figure 2 CT scan showing oval radiolucency surrounded by a well-defined radiopaque margin on the edentulous alveolar ridge with expansion on the buccal side.

higher magnification, the epithelium lining the cystic lumen was composed of ameloblast-like cells which were typically tall columnar in shape having palisading arrangement of nuclei with reversal of polarity. Stellate reticulum-like cells could also be seen on the superficial areas (figure 4). Few follicles within the connective tissue having peripheral columnar cells show palisading of nuclei, hyperchromatism and polarisation. The central cells within the follicles were stellate reticulum like in shape (figure 4).

The connective tissue wall of the cystic cavity was composed of dense fibrocollagenous stroma with few blood vessels and

Figure 3 Surgical tissue specimen with a whitish firm area.

extravasated red blood cells. Areas of haemorrhage were also evident. Few areas within the connective tissue wall showed multinucleated giant cells. The histopathological examination confirmed the lesion to be a unicystic ameloblastoma.

OUTCOME AND FOLLOW-UP

The patient was under regular follow-up till the healing of the surgical area was complete. The patient was last seen after a period of 3 months of the surgery with no recurrence. Currently, the patient has lost for the follow-up.

DISCUSSION

This report illustrates a case of residual cyst observed clinically and later confirmed to show features of both residual as well as ameloblastoma histopathologically. The residual cyst in this case is of prime concern as the epithelium of odontogenic cysts may be transformed into different tumours such as ameloblastoma,¹ adenomatoid odontogenic tumour² as well as squamous cell carcinoma.³

In 1977, Robinson and Martinez⁴ described a distinct variant of ameloblastoma on the basis of morphology, which they referred to as unicystic ameloblastoma. Lieder *et al*⁵ supported the findings of Robinson and Martinez and further hypothesised that the evolution of unicystic ameloblastoma may take place by either ameloblastic transformation of the reduced enamel epithelium of the developing tooth, or it may arise in an odontogenic cyst in which the neoplastic ameloblastic lining is preceded temporarily by a non-neoplastic stratified squamous

Figure 4 Photomicrograph showing a large cystic cavity lined by an arcading pattern of cystic lining transforming to ameloblastoma (top) and follicles of ameloblastoma within the connective tissue (bottom) (H&E stain, magnification $\times 200$, $\times 400$).

epithelial lining, or a solid tumour may undergo cystic degeneration of the ameloblastic islands with subsequent fusion of multiple microcysts to develop a unicystic lesion.

Ameloblastic transformation of common dentigerous, primordial, radicular and residual odontogenic cyst linings is the most plausible pathogenesis for cystic ameloblastomas.^{3 6 7} This was further supported by Lucas,⁸ who pointed out that a neoplastic transformation can occur in the epithelium of odontogenic cysts as both odontogenic cyst and ameloblastoma are of the same embryonic derivation.

Carpenter and Thoma⁹ and Small *et al*¹⁰ have reported one case each where an ameloblastoma was presumed to have arisen from a radicular cyst. Holmlund *et al*¹¹ reported two cases of radicular cyst enucleated and recurred as ameloblastoma later.

Thoma and Proctor¹² in 1937 described the case of a 31-year-old woman who apparently had an odontogenic cyst in the mandible, which on histological examination proved to be an ameloblastoma. Although the specific type of odontogenic cyst was not mentioned in this case, the description was consistent with a residual cyst of radicular origin. Lee¹ in 1967 reported a case of an ameloblastoma arising from a residual cyst in a 21-year-old female patient who presented with swelling in the left maxilla with a history of extraction of her permanent lateral incisor many years ago.

The present report is the second case to be reported in the literature illustrating an ameloblastoma arising from a residual cyst lining in a 62-year-old male patient, suggesting that such an ameloblastic transformation of the residual cyst can occur in the older age group as well and supporting the hypothesis that unicystic ameloblastoma is formed from the lining of an odontogenic cyst. The residual cyst in this case is of prime concern as the epithelium of odontogenic cysts may be transformed into different tumours such as ameloblastoma,¹ adenomatoid odontogenic tumour² as well as squamous cell carcinomas.³

The surgical approach to cystic lesions of the jaws is either marsupialisation or enucleation depending on the size and localisation of the lesion.¹³ Marsupialisation may well lead to a mistaken diagnosis, as the tissue removal in this procedure may be insufficient to reveal the tumour. Hence, in marsupialisation procedures, a large opening into the cystic cavity is needed not only to permit a larger area of the cyst contents for histological examination but also to provide adequate access for clinical examination of the cyst so that any suspicious areas of irregularity or mural thickening may be readily detected, in the presence of which complete enucleation of the cyst should be carried out. Unless this precaution is followed, it is possible that marsupialisation may create an erroneous impression that neoplastic change has occurred subsequently in the remaining portion of the 'cyst' wall when in fact the cyst so marsupialised was an *ameloblastoma* at the time of the surgery.

In the present case, fine-needle aspiration was carried out pre-operatively and a haemorrhagic fluid was obtained. Cytological examination showed surface epithelial cells of oral mucosa along with red blood cells and a clinical diagnosis of 'residual cyst' was made. On this basis, enucleation of the lesion was performed. For the present case, the earlier literature clearly indicated enucleation of the cyst as the preferred option, because marsupialisation carries the risk that any cystic epithelium left behind may become malignant.^{14 15}

Marsupialisation is favoured because of lower morbidity and it requires a long period of postoperative management. The major disadvantage of marsupialisation is that the epithelial

lining left in the cystic cavity may turn into a more aggressive lesion in the residual tissue. In most cases, later surgical enucleation will be necessary.^{16 17}

We believe that treatment decisions for ameloblastoma are based on the individual patient's situation. The surgical plan should be based on the clinicoradiological features, anatomic location, size of the lesion and age of the patient. Nevertheless, no matter what the diagnosis may be, the treatment option should be as conservative as possible.

Learning points

- This case illustrates the importance of subjecting all surgical specimens to histological examination, no matter how obvious the clinical diagnosis may be.
- Caries or trauma to the teeth can lead to development of radicular cyst, which in turn can lead to residual cyst, both of which have the potential to transform into ameloblastoma.
- A minimum 3–5 years of follow-up is necessary for every patient who is diagnosed with apical pathology.

Competing interests None.

Patient consent Obtained.

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