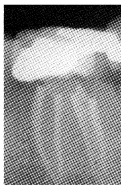

CASE REPORT

Central adenoid cystic carcinoma of the mandible manifesting as an endodontic lesion

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Abstract

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Aim To present a case of adenoid cystic carcinoma (ACC) in the mandible, and manifesting as a periapical lesion.

Summary A 56-year-old male suffered from pain around the right mandibular first molar for approximately 1 week. Oral examination revealed that the involved tooth was restored by a full coverage crown with no obvious abnormalities. A periapical radiograph revealed two ill-defined radiolucencies associated with the tooth, one over the mesial and another over the distal roots of the tooth; incomplete root filling and furcation involvement also being noted. The affected tooth was extracted based on the clinical impression of apical periodontitis. The surrounding tissue of the root apex was curetted and sent for histopathological examination, which revealed ACC.

Key learning points

- Adenoid cystic carcinoma affecting the mandible may mimic a periapical lesion. Proper diagnosis of such a lesion is dependent on thorough clinical, radiographic and microscopic examinations.
- Such a case highlights the benefits of biopsy and histological examination of collected tissues.
- Diagnosis of lesions in the mandible should include salivary gland tumours.

Keywords: adenoid cystic carcinoma, mandible, periapical, salivary gland tumour.

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Introduction

Adenoid cystic carcinoma (ACC), a malignant epithelial neoplasm of salivary gland origin, was originally described by Robin & Laboulbene in 1853 (Tauxe *et al.* 1962). The term

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'cyclindroma' was coined by Billroth (1859) due to the tumour's characteristic Swiss-cheese or cribriform appearance. From a review of ACC cases, around 30% of parotid, 30% of submandibular, and 40% of minor salivary-gland tumours, but only around 1% of sublingual-gland tumours are of this type (Cawson *et al.* 1998). ACCs arising centrally within the mandible would appear to be very rare, with, to the best of our knowledge, only about 20 such cases having been reported previously in the English-language medical literature (Bumsted 1955, Bradley 1968, Dhawan *et al.* 1970, Slavin & Mitchell 1971, Burkes 1975, Yoshimura *et al.* 1978, Kaneda *et al.* 1982, Gingell & Siegel 1983, Hirota & Osaki 1989, Johnson *et al.* 1989, Brookstone *et al.* 1990, Brookstone & Huvos 1992, Clark *et al.* 2000, Favia *et al.* 2000, Martinez-Madrigal *et al.* 2000, Abbott 2001). It is also noteworthy that three of the previously reported mandibular ACC cases occurred in the periapical region (Burkes 1975, Favia *et al.* 2000, Abbott 2001). This report presents a further case of mandibular ACC with clinical and radiological features mimicking a periapical lesion. The difficulties of accurate diagnosis and the potential for such a condition should constitute an alert to clinicians.

Case report

A 56-year-old male complained of pain around the mandibular right first molar for a period of about 1 week. He subsequently visited a local dentist and emergency periodontal treatment was performed on the provisional clinical diagnosis of periodontitis, although no radiographical examination was performed at that time. Following dental treatment, the patient returned home, but the symptoms recurred 2 days later, so he then presented at the Oral Pathology Department, Kaohsiung Medical University for further examination.

The patient revealed a history of cigarette smoking and alcoholism, although he denied betel-quid use. No other significant previous medical history was noted apart from diabetes mellitus controlled by insulin. The patient complained of spontaneous dull pain from the right mandibular first molar. Extra-oral examination revealed no facial asymmetry. Intra-oral examination revealed that the involved tooth was restored with a full coverage crown; nothing abnormal seeing was detected. The affected tooth was tender to percussion. There was no swelling of the alveolar ridge, nor any other abnormalities. In addition, the affected tooth was not mobile; periodontal probing was not performed. A periapical radiograph revealed two ill-defined radiolucencies (about 0.5 cm for the distal and 0.2 cm for the mesial root lesions), with no evidence of corticated bordering over the mesial and distal roots of the right mandibular first molar (Fig. 1). Incomplete root filling

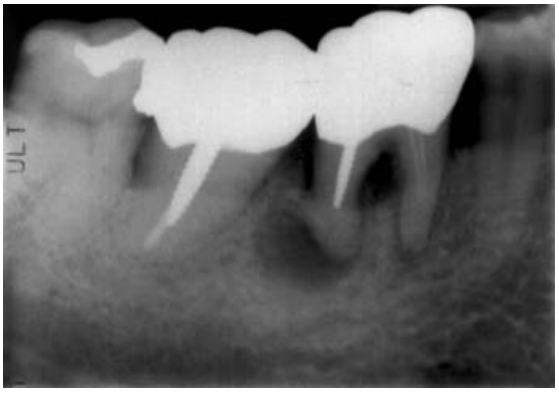

Figure 1 A periapical radiograph revealed two ill-defined radiolucencies with no corticated borders, located over the mesial and distal roots of the right mandibular first molar.

and furcation involvement were also noted (Fig. 1). In addition, a screw post was present in the distal canal with no apparent root filling in the apical region (Fig. 1). The provisional diagnosis was apical periodontitis.

The patient was referred to an endodontist for the possibility of removal of the crown and root canal re-treatment. The endodontist recommended tooth extraction because of the poor prognosis for endodontic re-treatment. Consequently, the affected tooth was extracted under local anaesthesia by an oral surgeon and the surrounding tissue of the root apex was curetted and sent for histopathological examination. During surgery, no communication to the right submandibular gland was noted.

Microscopic examination of the tissue fragments at low power revealed an epithelial malignant neoplasm with extensive infiltration of various differently sized tumour islands within the fibrous connective-tissue stroma (Fig. 2). At higher magnification, the tumour consisted of an irregular cluster in addition to sheets of polyhedral epithelial cells arranged in a predominant tubuloductal and cribriform pattern with cystic-containing hyaline or mucoid material (Fig. 3). However, some solid structures consisting of tumour cells were also noted (Fig. 3). Perineural infiltration was not evident. The morphological features of the tumour were consistent with the histological diagnosis of an ACC of salivary gland origin.

One week subsequent to tumour histopathology, the diagnosis of malignancy in the mandible was explained to the patient. Clinical examination revealed a healing post-extraction wound of the right mandibular molar; no regional palpable lymph nodes, and no other lesions in the major and minor salivary glands. The patient denied experiencing dyspnoea, dysphagia, and weight loss. Despite a thorough discussion with the patient concerning the histological findings, the patient refused treatment. The proposed treatment plan includes a wide mandibular surgical excision to remove any possible remnants of tumour cells, and further examinations including ultrasound and computerized tomography (CT) scanning of the salivary glands and cervical region. A whole-body CT scan and bone scintigraphy to exclude distant metastasis (lung and bone) was also proposed but rejected by the patient. Instead, the patient indicated his intention to visit a nearby teaching hospital for a second opinion and possible confirmation. As a consequence, the patient was unfortunately lost to follow-up.

Discussion

Salivary gland neoplasms affecting the jaw bone would appear to be very uncommon, with most cases being mucoepidermoid carcinomas (Brookstone & Huvos 1992). ACCs

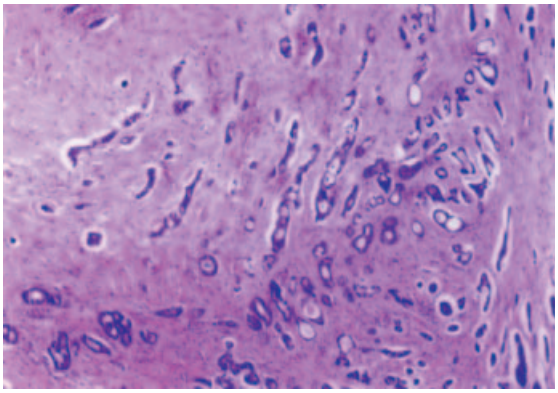

Figure 2 Low-power magnification (×40) reveals an extensive infiltration of varying-sized tumour islands within the fibrous connective tissue stroma of the tumour (H & E stain).

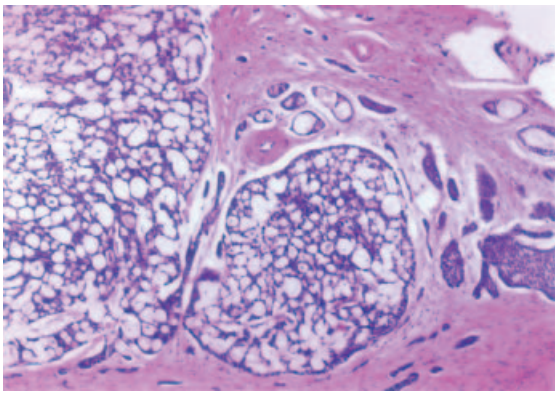

Figure 3 At a higher magnification (×100), the tumour consists of an irregular cluster and also sheets of polyhedral epithelial cells arranged in a predominant tubuloductal and cribriform pattern with associated pseudocystic hyaline or mucoid bodies (H & E stain).

Table 1 Diagnostic criteria for primary intra-osseous salivary gland neoplasms (Batsakis 1979)

Diagnostic criteria
1. Radiographical evidence of osteolysis
2. Presence of intact cortical plates
3. Presence of an intact mucous membrane overlying the lesion
4. Absence of any primary tumour within the major or minor salivary glands
5. Histopathological confirmation of the typical architectural and morphological features of a salivary gland tumour

occurring within the periapical region would also appear to be extremely rare (Brookstone & Huvos 1992).

Radiographically, most reports pertaining to intra-osseous salivary gland tumours appear to describe their presentation as unilocular or multilocular osteolytic radiolucencies (Brookstone & Huvos 1992). For this case, a unilocular radiolucency located in a periapical area was observed. In addition, local pain appears to be a common finding for such lesions (Brookstone & Huvos 1992), as in this case. It can be misinterpreted as of dental origin and due to incomplete endodontic treatment of the involved tooth. The correct diagnosis was only derived from the histological examination of the biopsied tissue. Consequently, the present case represents an intra-osseous ACC masquerading as a lesion of apical infection indicating that a proper diagnosis of such a lesion is dependent not only upon thorough clinical and radiographical examinations, but also upon the accurate interpretation of biopsied material. Furthermore, this particular case highlights the importance of the detailed histological examination of any tissues taken from the periapical region. In addition, central salivary gland tumours should be included in the differential diagnosis of lesions in the mandible, particularly for those located in the periapical area.

The diagnostic criteria for primary intra-osseous salivary gland neoplasms have been proposed previously (Batsakis 1979) (Table 1). For the case reported here, all but the fourth diagnostic criteria were satisfied, as the patient refused an extensive work-up and examination.

The pathogenesis of intra-osseous salivary gland tumours remains controversial and several theories explaining the possible mechanisms for these tumours have been described (Browand & Waldron 1975, Bruner & Batsakis 1991) (Table 2) but no single theory of histogenesis is applicable to all cases of central salivary gland tumours. True

Table 2 Possible mechanisms of intra-osseous salivary gland tumours (Browand & Waldron 1975, Bruner & Batsakis 1991)

Possible mechanisms
1. Ectopic salivary gland tissue resulting from the entrapped minor salivary glands, or inclusions of embryonic rests of submandibular and sublingual glands or seromucous glands displaced from the maxillary sinus into the maxilla
2. Neoplastic transformation of the mucous-secreting cells commonly found in the epithelial lining of odontogenic cysts

cases of central ACC do not arise from a metaplasia of odontogenic cysts and therefore, quite possibly, originate from the ectopic salivary gland tissues (Brookstone & Huvos 1992).

Brookstone & Huvos (1992) reported that salivary gland neoplasms arising within the boundary of the jaw bones could behave differently to conventional salivary gland tumours found in the major and minor salivary glands, and therefore proposed a new clinical staging I–III for such lesions. The present case can be categorized as being a stage I disease, which has a fairly good prognosis.

Intra-osseous salivary gland tumours do not differ microscopically from their soft-tissue counterparts. Histologically, there appear to be three recognizable patterns of growth: solid, cribriform and tubuloductal.

The main treatment for central ACC is excisional surgery (Brookstone & Huvos 1992). A wide surgical resection paying particular attention to obtain clear margins around regional nerves was the suggested treatment as this tumour demonstrates a propensity for perineural growth. Furthermore, for such a condition, long-term follow-up is essential because this tumour type is notorious for its late and persistent recurrence and distant metastases, most commonly to the lung, brain and bone, in nearly 70% of patients at the time of their death (Stell 1986).

In conclusion, central ACCs of the mandible occurring in the periapical area are rare, and they are a diagnostic challenge to every clinician in that they may easily be confused with lesions of endodontic origin.

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