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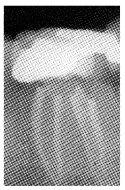

CASE REPORT

Intraosseous schwannoma mimicking a periapical lesion on the adjacent tooth: case report

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Abstract

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Aim To present an additional case of intraosseous schawannoma involving the apical area in the mandibular alveolar bone mimicking an inflammatory periapical lesion.

Summary This article describes a case of schwannoma periapically located mimicking an inflammatory periapical lesion in the mandible of a 34-year-old female. Diagnostic and therapeutic problems can occur when this lesion is misinterpreted as being endodontic in origin. The diagnosis, radiograph, immunohistochemical aspects and treatment are also discussed.

Key learning points

- Intraosseous schwannoma is a rare unilocular radiolucency that when located periapically could be misdiagnosed as an endodontic lesion and result in unnecessary root canal treatment.
- The vitality of the pulp is an important test to exclude lesions of inflammatory origin.
- Histological examination is important to establish the diagnosis of lesions in the periradicular region.

Keywords: benign tumour, mandible, neural tumour, neurilemmoma, S100, Schwann cells, schwannoma.

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Introduction

Schwannomas were first established as a pathologic entity in 1910 by Verocay who called them neurinomas (Verocay 1910). The terms schwannoma or neurilemmoma presently

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describe solitary encapsulated nerve tumours of ectodermal origin arising from perineural Schwann cells of the nerve (Hatziotis & Asprides 1967, Zachariades *et al.* 1987).

Schwannomas are slowly growing asymptomatic tumours that have a predilection for the head, neck and flexor surface of the upper and lower extremities (Hatziotis & Asprides 1967, Zachariades *et al.* 1987), occurring most commonly in the tongue followed by the palate, floor of the mouth, oral mucosa, gingivae, lip and buccal mucosa (Verocay 1910, Wright & Jackson 1980).

Intraosseous schwannomas are rare, representing <1% of benign primary tumours of the bones. The radiographical features of these lesions are nonpathognomonic, that makes their diagnosis difficult. The site most commonly involved is the mandible and sacrum. Intraosseous schwannoma of the maxilla is exceptionally unusual (Marzola *et al.* 1988, Llewelyn & Sugar 1989, Takeda 1991, Minic 1992, Villaneuva *et al.* 1995). Recent studies have shown a substantial increase in the number of intraosseous schwannomas in the recent years and a preponderance for women and patients in the second, third and fourth decades (Hatziotis & Asprides 1967, Zachariades *et al.* 1987, Takeda 1991). When localized close to the periapical region or laterally on roots these lesions could be confused with inflammatory odontogenic lesions.

The malignant transformation of schwannoma is extremely rare as opposed to the transformation of neurofibroma in connection with neurofibromatosis (Enzinger & Weiss 1988).

The aim of this report is to present a clinical case of schwannoma in the mandibular alveolar bone mimicking an inflammatory periapical lesion. The radiographic, histological, immunohistochemical and treatment aspects are emphasized. The report discusses the importance of differential diagnosis and complementary examinations such as pulp vitality tests and histological examination to exclude inflammatory lesions and obtain the diagnosis.

Case report

A 34-year-old white female was referred to the School of Dentistry of Nove de Julho University, São Paulo, Brazil complaining of pain in the mandibular right molar region for more than a week. This pain was made worse with cold food and drinks. Clinical examination revealed no expansion of the mandible bone. The overlying mucosa was intact and of normal colour. Radiographical examination revealed a radiolucent area under a composite restoration in the first molar indicating a carious lesion. In the alveolar bone, a radiolucent, unilocular, well-circumscribed lesion with a dense sclerotic border situated between the second premolar and first molar in the right mandibular area was noted. The panoramic radiograph showed that the lesion was a single area of pathosis with a size of 1.5 × 1.0 cm in its greatest diameters. The periodontal attachment and lamina dura of both teeth appeared to be intact (Figs 1 and 2). The second premolar and first right molar teeth responded to pulp sensitivity tests. The patient was unaware of the lesion and did not know for how long it had been present. The remainder of the history was not contributory. The differential diagnosis was some type of odontogenic lesion such as a keratocyst or ameloblastoma, a central giant cell granuloma or idiopathic bone cavity.

After local anaesthesia, the lesion was excised surgically. During surgery, the lesion appeared well-circumscribed, unilocular, greyish white in colour and was situated between the roots of the first molar and second premolar teeth. It was easily separated from the surrounding tissue.

The old composite restoration was removed and an indirect pulp capping procedure was undertaken.

Microscopic examination of the histological sections revealed an ovoid piece of tissue surrounded by a thin fibrous connective tissue capsule. The lesion was composed of

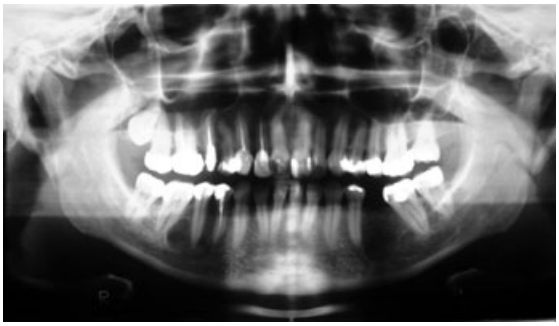

Figure 1 Panoramic radiograph showing the lesion between the right second mandibular premolar and first molar.

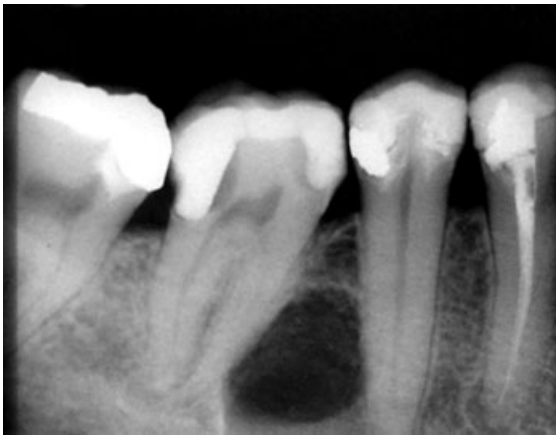

Figure 2 Periapical radiograph revealing a well-circumscribed radiolucent image in alveolar bone with a dense sclerotic border.

tissue, which had a definite neural pattern. Antoni type B tissue aspect was predominant. The lesion was composed of densely cellular and irregularly arranged, elongated spindle tumour cells (Fig. 3). Numerous large, thin-walled blood vessels were also present; these became more numerous closer to the centre of the lesion. It was not possible to identify a peripheral nerve related to this tumour.

On the basis of its classic histological features, the diagnosis was benign schwannoma. Immunohistochemical examination showed that the tissue stained readily for S100 protein confirming the histological examination (Fig. 4).

The postoperative course was uneventful and healing was normal. There were no signs of recurrence 3 years later. During this period the patient had more pulp pain and root canal treatment in the molar tooth was completed. The radiograph revealed complete bone formation in the surgical site (Fig. 5). Based on the diagnosis, a general physical examination was completed to rule out multiple tumours of neural origin. The results of this examination were negative.

Discussion

This paper reports an intra-bony schwannoma arising in the mandible adjacent to tooth roots. Schwannoma, also called neurilemmoma, is an infrequent, usually benign tumour,

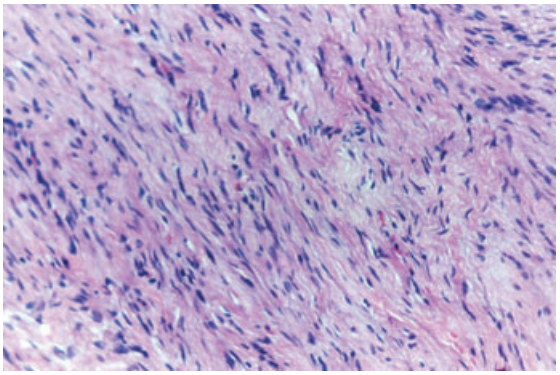

Figure 3 Photomicrograph of the lesion composed of densely cellular and irregularity arranged, elongated spindle tumour cells. Antoni B tissue was dominant. The tumour cells had fusiform aspect with no precise cytoplasmic limits (H&E ×100).

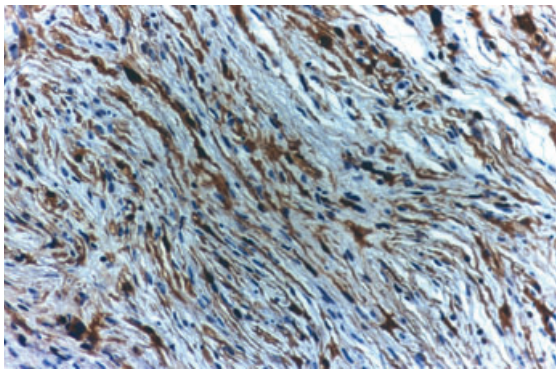

Figure 4 S100 positive reaction in schwannoma. Immunoperoxidase staining (×400).

derived from the neuroectodermal Schwann cells that cover myelinated nerve fibres (Hatziotis & Asprides 1967, Zachariades *et al.* 1987). The neoplasm has a predilection for the head, neck and flexor surfaces of upper and lower extremities. Whereas the head and neck region is the most common site for benign nerve sheath tumours, intra-oral lesions are unusual and only a small number occur in intraosseous sites (Marzola *et al.* 1988, Llewelyn & Sugar 1989, Takeda 1991, Minic 1992, Villaneuva *et al.* 1995).

Intraosseous lesions located in the periapical region could lead to clinical and radiographical diagnostic confusion. Most of the lesions situated at these sites are odontogenic in origin. However, other possibilities must be included in the differential diagnosis, especially when the dental lamina are preserved and pulp tests are positive.

The literature reveals 44 reported central schwannomas of the jaws; 39 in the mandible and 5 in the maxilla (Chi *et al.* 2003). Usually, this lesion exhibits a behaviour similar to other benign intraosseous lesions. Based on this slow growth the radiographical features include a well-defined radiolucent area, which may be bordered by a dense sclerotic margin of bone. The expansive pattern of growth tends to dislocate involved nerves and teeth (Marzola *et al.* 1988, Llewelyn & Sugar 1989, Takeda 1991, Minic 1992, Villaneuva *et al.* 1995).

The present report demonstrates a case of intraosseous schwannoma that was diagnosed following meticulous examination by the dental surgeon. The patient

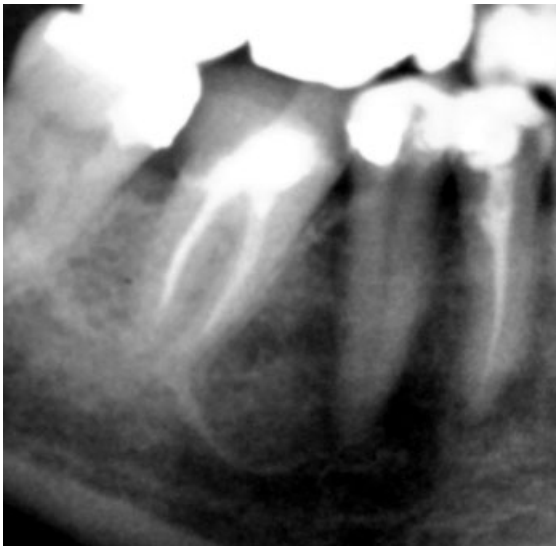

Figure 5 Closer view of panoramic radiograph 1 year after the complete excision showing bone regeneration in the area.

complained of acute toothache and a radiographical examination was carried out to evaluate the tooth and restoration. Simultaneously a well-circumscribed radiolucent area between the second premolar and first molar roots was observed. The lesion could be confused with inflammatory periodontal lesion such as dental granuloma or radicular cyst, although clinical findings seemed to indicate a nonendodontic origin. The involved tooth gave a positive result to sensitivity tests. An exploratory surgical (curettage) approach was chosen, and the final diagnosis of the tissue mass was microscopically confirmed to be a schwannoma.

The microscopic analysis of the specimens obtained is extremely important to establish the final diagnosis. Some clinicians do not routinely send the specimens for histological examination, this can lead to erroneous diagnosis and treatment. The intraosseous schwannoma is a rare lesion and the clinical course and the radiographic appearance of this lesion is not characteristic; therefore, the diagnosis can only be established on histological examination (Fiedman 1964, Hatziotis & Asprides 1967, Schofield & Gadner 1982, Gotte *et al.* 1986, Gallo *et al.* 1997). The extent of the lesion at diagnosis may indicate more radical procedures, including removal of teeth and bone resection (Marks *et al.* 1976, Llewelyn & Sugar 1989, Gallo *et al.* 1997).

When seen as a gross specimen, the tissue of schwannoma is solid, roundly lobulated and greyish white in colour. It is soft or moderately firm. Most tumours are encapsulated. Microscopically, the growth is classified into two basic types of tissue, Antoni A and Antoni B. The Antoni A type consists of closely packed spindle cells with their nuclei lying in rows and displaying a palisade effect. Hyaline structures areas are observed between these rows; those with round to oval configuration are called Verocay bodies. The Antoni B type of tissue is composed of a loose-meshed cells haphazardly arranged with number of minute cystic spaces (Lucas 1984, Zachariades *et al.* 1987, Enzinger & Weiss 1988, Chrysomali *et al.* 1997).

According to Chrysomali *et al.* (1997) in terms of immunohistochemistry, all neural origin tumours have positivity for S100 protein and this technique could help distinguish these lesions. Intense positive reaction for S100 is observed in schwannoma and

palisaded encapsulated neuroma. Intensive reaction for CD57 is observed in traumatic neuroma, capsular EMA (epithelial membrane antigen) and CD34 stainings are observed in schwannoma.

The histopathological diagnosis of a biopsy may present difficulties in establishing the true nature of the tumour, as malignant tumours of neural origin are characterized by many of the same features as benign nerve sheath tumours (Enzinger & Weiss 1988). On the other hand, especially in these tumours, it is essential to ascertain whether it is benign or malignant. The treatment of malignant lesions is radical resection, whilst the benign forms merely need extirpation (Colmenero *et al.* 1991). Disturbances of nerve function are not a clinical criteria for distinguishing the behaviour of the tumour, although the malignant neoplasm tends to cause more pain in the region of distribution of the nerve (Krause *et al.* 1993).

The treatment for schwannoma consists of total surgical lesion removal. In the present case, the surgical exploration was completed and the lesion was totally excised with no signs of recurrence 3 years later. Recurrences and malignant transformation are rare events. The presence of schwannoma must lead to a careful search for neural tumours in other parts of the body, although in most cases none will be found. The differentiation of schwannoma from neurofibroma is essential, because an apparently 'solitary' neurofibroma may be a manifestation of neurofibromatosis.

Conclusion

Introsseous schawannoma are rare lesions and can mimic periapical lesions. Careful diagnostic procedures and adequate interpretation of clinical, radiographical and histological findings of intraosseous lesions situated at periapical region are required to associate the correct diagnosis, which otherwise may result in unnecessary root canal treatment.

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