Schwannoma Located in the Upper Gingival Mucosa: Case Report and Literature Review

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Abstract The Schwannoma is a benign tumor that originates from Schwann cells of peripheral motor and sensory nerves. It presents as a solitary, slow growing, smooth-surfaced and asymptomatic firm mass. The pre-operative diagnosis is often difficult, and in majority of cases it can only be made during the surgery and the definitive diagnosis obtained by histological study when cells test positive for S-100 protein. In this report, a case of a 20-year-old female patient with a Schwannoma located in the upper gingival mucosa is discussed. The patient was referred to the department of oral and maxillofacial surgery with chief complaint of enlargement of the upper gingival mucosa at the level of upper right premolar. Clinical findings showed characteristics of connective tissue tumor including epulis; however, histologic and immunohistochemical studies revealed it to be a Schwannoma. There has been no sign of recurrence 1 year and 8 months after surgery.

Keywords Schwannoma · Gingival mucosa · Soft tissue tumors · S-100 protein

Introduction

Schwannoma (also known as neurilemmoma) is a benign nerve sheath tumor arising from perineural Schwann cells, which develop during the 4th week of gestation when the ectomesenchymal detachment of the neural crest occurs [1–3]. In addition to typical Schwannoma with classic Antoni A and Antoni B areas, many variant types have been described, such as plexiform, cellular, epithelioid, and ancient Schwannomas [4].

The tumor is frequently located in the head and neck region (about 25–45 %), but only few (1 %) have an intraoral origin showing a predilection for the mobile portion of the tongue; however, Schwannoma of the gingival mucosa is extremely rare [1, 2, 5–7].

Case Report

A 20-year-old female was referred with a history of a slowly growing, symptom-free mass at upper right premolar gingiva since 6 years. Medical history was not contributory.

On clinical examination, an elastic-soft, well-circumscribed nodule measuring approximately $25 \times 15 \times 7$ mm covered with normal oral mucosa on the lower part was noted (Fig. 1). The mass was tightly fixed to the buccal alveolus and was immovable. No inflammatory changes or other alterations were found in the surrounding tissues. The upper right premolar responded positively to pulp testing with no tooth mobility. A dental radiograph showed no particular findings (Fig. 2).

A resection of the tumor was performed under local anaesthesia (lidocaine hydrochloride 2 % with adrenaline 1:80,000). The tumor was resected with surrounding

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Fig. 1 Clinical examination of tumor mass revealed lesion located in the right gingival mucosa at level of upper right premolar

Fig. 2 The alveolar bone was intact on radiographic examination

gingival mucosa and periosteum for pathological examination. Under the resected well-encapsulated mass, was observed partial resorption of the buccal alveolar bone of the upper right premolar (Fig. 3a). A collagen sponge (Terudermis®, Olympus Terumo Biomaterials Co. Ltd., Tokyo, Japan) was used to promote epithelialization (Fig. 3b).

Microscopically, hematoxylin and eosin stained sections revealed proliferation of spindle cells organized in a palisaded pattern (Antoni A) with myxoid degeneration (Antoni B) (Fig. 4a–c). Atypical or bizarre cells with large

nuclei were often observed but mitotic figures were scantily seen. Immunohistochemical examination of S-100 protein and positive reactivity was observed in both the cytoplasm and nuclei of the neoplastic cells (Fig. 4d). These findings were compatible with the diagnosis of Schwannoma. There has been no sign of recurrence 1 year and 8 months after surgery (Fig. 5).

Discussion

Schwannomas often occur in the head and neck region with a majority in the carotid triangle [8]. Intraorally, the tongue is the most common location, with a predilection for the anterior portion of the palate. Floor of the mouth, the cheek mucosa, the lip and the gingivae are also affected, in order of decreasing occurrence [9]. A review from 1960 to 2006 revealed four cases and, a Japanese review from 1976 to 1999 had reported only one case on gingival mucosa [10, 11]. Moreover, from 2001 to 2011, we observed four cases of Schwannoma in our clinic; three on the tongue, one arisen from the gingival mucosa. This result supported previous studies that the intraoral Schwannoma seems to be rare in clinical practice.

Clinically, most oral Schwannomas are asymptomatic, like in our patient, and some are discovered during routine examinations. The tumor is typically solitary and slow growing. Also, ulceration is occasionally seen, and it is probably due to trauma [12].

The initial diagnosis is difficult because of rarity of this condition and the lack of specific symptoms and signs associated with these tumors. Differential diagnosis (by means of histological study) includes malignant tumors, inflammatory and cystic lesions, benign epithelial and connective tissue tumors. In our case differential diagnosis was also made with lipoma, and epulis.

Microscopically, Schwannoma consist of a thin fibrous capsule and a tumoral proliferation commonly formed by two patterns, Antoni A and Antoni B. The first are formed by fusiform cells with elongated nuclei arranged in a well organized palisading pattern. Antoni B shows a disordered arrangement of cells distributed in a loose fibrillar matrix with areas described as microcysts. The immunohistochemical tests reveal the Schwannoma cells to be positive for the protein S-100, a marker for the nervous system. This test is expressed by Schwann cells, but is not expressed by perineural cells or endoneurial fibroblasts [13, 14].

The treatment of choice is surgical excision. The encapsulated form is enucleated easily, whereas the non-encapsulated requires normal tissue margins to avoid relapse. If the nerve of origin is visualized, an attempt should be made to separate it carefully to preserve

Fig. 3 The tumor was well-encapsulated and totally excised (**a**). A collagen sponge was used (**b**) to support the free bone flap of the alveolar bone and to promote epithelialization of the gingival mucosa

Fig. 4 **a** The tumor was compounded from Antoni A and B areas (H and E, $\times 20$). In **b** nuclei are arranged in palisades separated by ribbed collagen (Antoni A, $\times 40$), while in **c** there is some disordered

arrangement of cells distributed in a loose fibrillar matrix (Antoni B, $\times 40$). **d** Immunohistochemical staining of S-100 protein indicated that the tumor cells were strongly positive ($\times 40$)

function. In our case the connection with the nerve could not be seen; as occurs in other locations of the oral cavity such as the upper lip [15].

The prognosis is very good especially on intraoral Schwannoma, when the tumor is extirpated in its entirety, and malignant transformation is rare. Although, some

Fig. 5 After 1 year and half, there is no sign of recurrence ($\times 40$)

reports showed malignancy transformation with parotid and lung metastasis, and; constant post-operative follow-up is needed [16, 17].

Conclusions

A case of Schwannoma located in the upper gingival mucosa was reported. Schwannomas should not be discarded when observing a well-circumscribed mucosal tumor in the oral cavity. The clinical examination is the key for the suspected tumor, and the biopsy with anatomopathologic study should be carried out for a definitive diagnosis.

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