



CASE REPORT

Ancient schwannoma of the mouth floor – A case report and review

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KEYWORDS

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Summary Ancient schwannoma rarely occurs in the oral cavity. Reviewing the literature, only six intra-oral cases of ancient schwannoma have been reported in the English-language medical literature. Here we present an intra-oral ancient schwannoma in a male patient with, to our knowledge, the longest reported duration: 18 years. In addition, a brief review of the pertinent literature is included.

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Introduction

Schwannoma (neurilemmoma) is an uncommon, benign, encapsulated tumor arising from nerve sheath cells.¹ Histopathologically, five schwannoma variants have been described: common, plexiform, cellular, epithelioid, and ancient schwannomas.²

Ackerman and Taylor (1951)³ reviewed 48 cases of thoracic tumors of neurogenic origin and found 10 cases that showed features consistent with common schwannoma, with distinctive areas of hypocellular tissue. They attributed these features to long-standing degenerative alterations, and coined the term 'ancient' for this type of benign neurogenic tumor. Since that first report, ancient schwannoma has been described in a variety of locations including the oral cavity.^{4–9}

To our knowledge there have only been six cases of ancient schwannoma reported in the oral cavity in the English-language medical literature (Table 1).^{4–9} Here, a case of an ancient schwannoma of the mouth floor with some

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Table 1 Reported cases of ancient schwannoma in the oral cavity

Authors (year reported)	Sex	Age (yrs)	Site	Largest diameter (cm)	Duration (yrs)
Eversole & Howell (1971)	Female	58	Mouth floor and ventral tongue	2.5	Not provided
Marks et al. (1976)	Female	65	Mouth floor, right	3.5	Not provided
McCoy et al. (1983)	Female	36	Maxillary left posterior mucobuccal fold	2.0	Not provided
Dayan et al. (1989)	Female	52	Upper left vestibule	0.9	Not provided
Nakayama et al. (1996)	Female	40	Mouth floor and ventral tongue	5.5	2 months
Ledesma et al. (1999)	Female	21	Mouth floor and ventral tongue	3	5 months
Chen et al. (present case)	Male	34	Mouth floor, left	3	18 years

unreported features is presented and the literature on intra-oral schwannomas is reviewed.

Case report

A 34 year-old male patient, with no symptoms or family history of von Recklinghausen's disease, attended our institution with the chief complaint of an asymptomatic, firm, 18-year duration swelling, measuring about 3 cm in maximum diameter, over the left anterior mouth floor (Fig. 1). The patient stated that the size of this swelling had remained unchanged during this 18-year period. Laboratory tests and clinical history were non-contributory. The clinical impression was of a benign tumor of mesenchymal origin. Under general anesthesia, a well encapsulated tumor was removed with blunt dissection. The surgical specimen was sent to the Oral Pathology Department of our institution for histological examination. The patient's recovery was uneventful and he was discharged one week post-surgery. During two years of review, the



Figure 1 A well-circumscribed swelling over the left anterior mouth floor.

patient showed no clinical or radiographic evidence of recurrence.

The resected specimen consisted of a well-circumscribed mass measuring $3 \times 3 \times 2.5$ cm (Fig. 2A). The cut surface showed both solid and cystic areas (Fig. 2B). Microscopic examination revealed an encapsulated tumor with solid cellular areas, hypocellular areas and a large cystic space (Fig. 3A). At the periphery of the mass, cells consisting of proliferating groups of palisading nuclei, consistent with Schwann cells (Figs. 3B and C), forming Verocay bodies were observed (Fig. 3D). In some areas, the tumor was infiltrated by large numbers of siderophages and histiocytes (Figs. 4A and B). Degenerative changes (Fig. 3A and B) and hyalinization (Fig. 4C) were also seen. Blood vessels of various sizes were also noted. Immunohistochemical investigation of the tumor cells showed diffuse, strongly positive staining for S-100 protein (Fig. 4D). According to these histological findings, the diagnosis of an ancient schwannoma of the left mouth floor was made.

Discussion

Approximately 25–40% of all schwannomas occur in the head and neck area¹⁰ whilst ancient schwannoma rarely affects the head and neck region,¹¹ especially the oral cavity. The literature review yielded 22 well-documented cases of ancient schwannoma reported in the head and neck region¹² in which only six cases were in the oral cavity (Table 1).^{4–9}

Eversole and Howell (1971)⁴ reported the first intra-oral case of ancient schwannoma, although Cherrick and Eversole¹³ presented two additional cases in the same year but detailed clinical information was not given. In the present report, an additional case with some new features is described. Reviewing the literature revealed that all cases of oral ancient schwannoma were in females,^{4–9} unlike this case. Furthermore, our case is of interest because it is has the longest reported duration of 18 years.

Ancient schwannoma cannot be clinically distinguished from other types of schwannoma. The mean age of occurrence for the seven patients now reported is 43.7 years (range, 21–65 years). The clinical appearance of the lesions

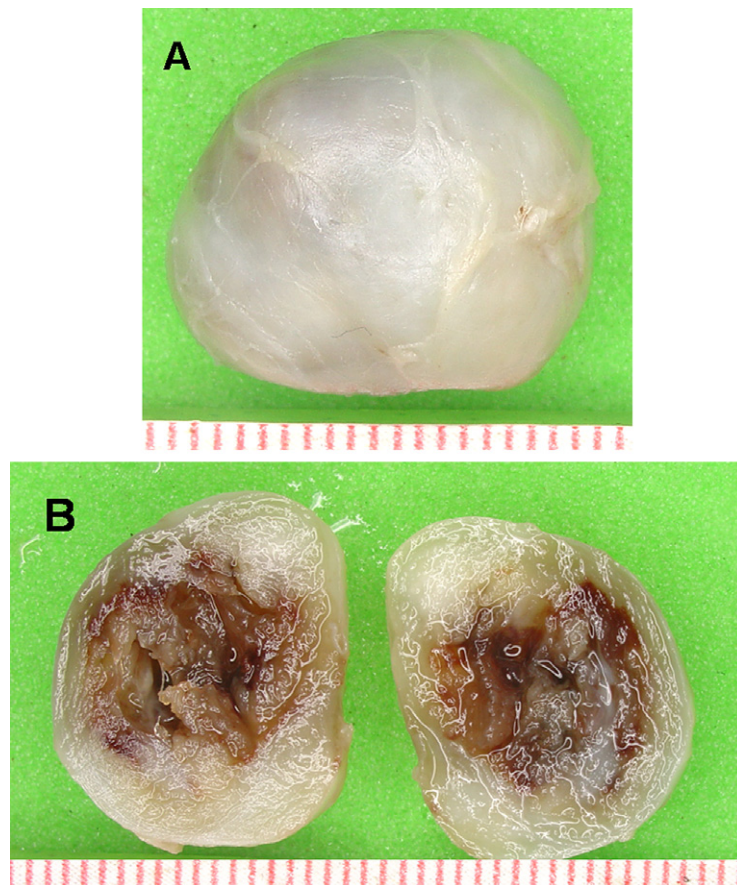


Figure 2 Photograph of the surgical specimen (A). The cut surface shows a well-encapsulated tumor mass containing both solid and cystic areas (B).

was of submucosal swellings of diameter between 0.9 and 5.5 cm. All the intraoral lesions, including our case, were well circumscribed and demarcated from the surrounding tissues.^{4–9}

When common schwannomas are present in the oral cavity, they are most often in the tongue.¹³ From our review of the literature, most intra-oral ancient schwannomas (5 cases including our own) were located in the mouth floor;^{4–7} in two cases the ventral tongue was also involved.^{8,9} Only three cases, including the current case, documented the duration of the lesion (2 months, 3 months and 18 years respectively).^{8,9} No recurrence has been noted to date for any of the reported intraoral cases.^{4–9} In addition, no malignant transformation occurred in any of the reported intraoral ancient schwannomas, suggesting that the treatment of choice is surgical excision.^{4–9} This finding suggests that ancient schwannoma is not an aggressive neoplasm. However, it should be carefully noted that malignant transformation has been reported in some cases of ancient schwannoma from other locations.¹⁴

Ancient schwannomas can vary from firm, solid masses to fluctuant cysts. The predominant histopathological findings are of an encapsulated lesion consisting of a mixture of spindle cells forming highly cellular so-called Antoni A tissues and less cellular, myxoid Antoni B tissues.⁴ Characteristically, this tumor is long-standing and may undergo degenerative 'ancient' changes dominated by large cystic, myxoid areas with variable bizarre spindle cells and occasional mitoses.¹⁵ In the case described here, the long-standing tumor (18 years) was encapsulated, and consisted of both solid and cystic areas as observed in the previous reports. However, the histological diagnosis in the present case presented some difficulties since only the peripheral part of the lesion was of significant value in the diagnosis of its neurogenic origin. This consisted of palisaded Schwann cells in Antoni A tissue and was confirmed using immunohistochemistry.

In conclusion, we present an intra-oral ancient schwannoma which, to our knowledge, is the first such report in a male patient and has the longest reported duration (18 years).

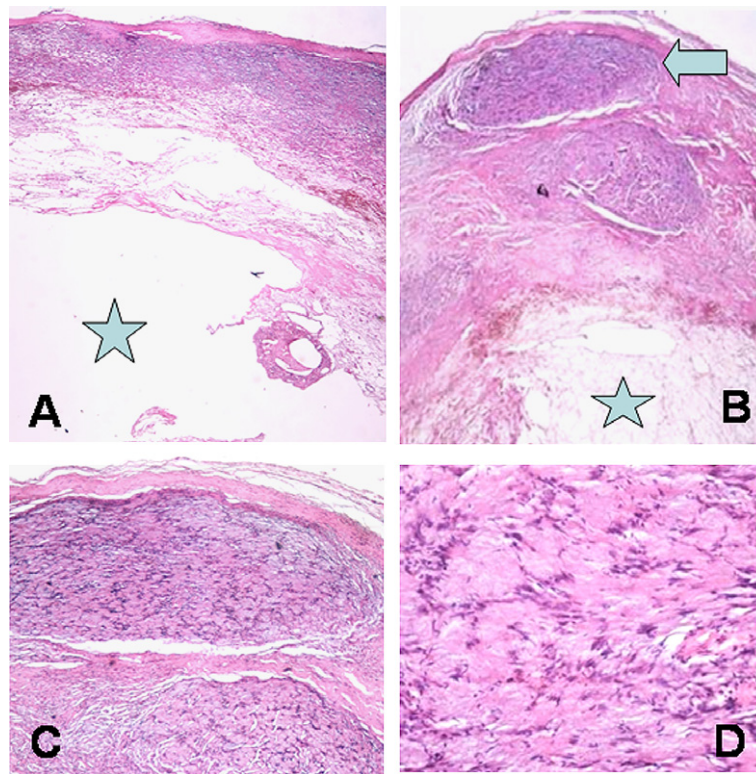


Figure 3 Hematoxylin and eosin staining of the specimen showing an encapsulated tumor with solid cellular areas, hypocellular areas and a large cystic space (as indicated by the star) (A & B, $\times 40$). Tumor cells composed of proliferating groups of palisaded Schwann cell nuclei in Antoni A tissue (B, $\times 40$ & C, $\times 100$) forming Verocay bodies (D, $\times 100$) were noted at the periphery of the mass.

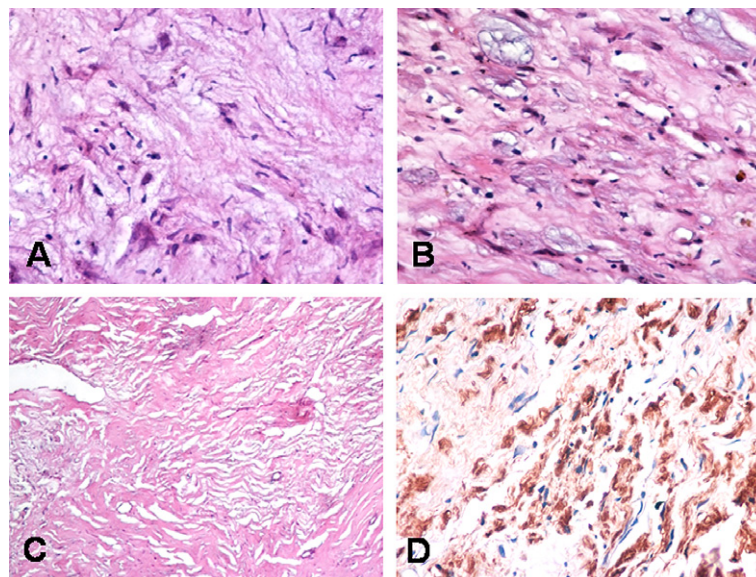


Figure 4 Hematoxylin and eosin staining of the tumor showing infiltration of siderophages and histiocytes in some areas (A & B, $\times 100$). Degenerative changes (A & B, $\times 100$) and hyalinization (C, $\times 40$) were also seen. Immunohistochemical staining of the tumor cells revealed diffuse, strongly positive staining for S-100 protein (D, $\times 100$).

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