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Case Report

Ancient Schwannoma of oral cavity in a pediatric patient

Manas Bajpai, Nilesh Pardhe, Manish Kumar*

Department of Oral and Maxillofacial Pathology, NIMS Dental College, Jaipur, India
* S.P Medical College, Bikaner, India.

Abstract Schwannomas are benign tumors of peripheral nerve sheath origin, composed of proliferation of Schwann cells in a characteristic pattern. Ancient schwannomas are long-standing lesions exhibit degenerative changes and atypia due to which they could be mistaken for malignancy. The mean age of occurrence of AS in oral cavity is 43 years and no case of AS has been reported in a pediatric patient so far. Here we present a rare case of AS of oral cavity in a 10 year old male child.

Key words
Schwannoma, ancient schwannoma, peripheral nerve sheath tumor, pediatric patient.

Introduction

Oral schwannomas are uncommon tumors comprising of 1% of all schwannomas, they may arise both in soft tissue and bone.[1] Those arise in soft tissues may mimic other lesions such as, mucocele, neurofibroma, fibroma, lipoma, peripheral ossifying fibroma, peripheral giant cell granuloma, pyogenic granuloma etc.[2] Ancient schwannoma (AS) is considered as a long-standing variant of schwannoma, histologically characterized by hemorrhage, hemosiderin pigmentation and pleomorphism of cells.[3] AS arising in oral cavity is exceedingly rare and only 18 cases have been reported in the literature as per our best knowledge.[4] An exhaustive review of literature did not reveal a single case in a pediatric patient. A case of AS in a pediatric patient arising in buccal mucosa is presented here.

Case report

Address for correspondence
Dr, Manas Bajpai
Department of Oral and Maxillofacial Pathology,
NIMS Dental College,
Jaipur, India
Email: dr.manasbajpai@gmail.com

A 10-year-old male child presented to a private clinic in Jaipur (India) by his parents, for the evaluation of an asymptomatic swelling on his upper, front region of the jaw since 10 months. Past medical and family history of the patient was not relevant to the present swelling. Past dental history revealed a scaling of teeth 2 years back in government hospital. Intraoral examination revealed a soft and fluctuant swelling on the lingual surface of maxillary gingiva between #11 and #21 measuring about 2 × 2 cm in diameter. The color of the swelling was reddish but no discharge was observed **(Figure 1a)**. An intraoral periapical radiograph was taken that showed no relation of swelling with the bone. A provisional diagnosis of pyogenic granuloma was made. The lesion was surgically excised and tissue was sent to the Department of Oral and Maxillofacial Pathology, NIMS Dental College Jaipur (India) for expert opinion **(Figure 1b)**.

Histopathological evaluation of the specimen revealed a circumscribed mass comprised of spindle-shaped cells arranged in Antoni A configuration surrounding eosinophilic structures looked like verocay bodies **(Figure**

2a). Other cellular pattern showed an irregular arrangement of spindle cells without palisading, suggestive of Antoni B configuration. Stroma was vascular and made up of numerous dilated blood vessels; a large thrombus was noted in the centre with hemosiderin pigmentation **(Figure 2b)**. Focal area of myxoid degeneration was seen and few areas showed hemorrhage with the collection of numerous extravasated RBCs. Spindle cells showed some atypical features like cellular pleomorphism and nuclear hyperchromatism. Based on all the features, final diagnosis of ancient schwannoma rendered.

The follow-up period of 6 months was uneventful.

Discussion

Oral schwannoma is a rare benign, slow growing, asymptomatic tumor of neural origin.[2] Term AS was coined by Ackerman and Tayler in 1951 to long-standing schwannoma exhibited degenerative changes.[5] Oral AS was first reported in 1971 by Eversole and Howell.[6] Only 18 cases of oral AS have been reported in the English language medical literature till date.[4] All

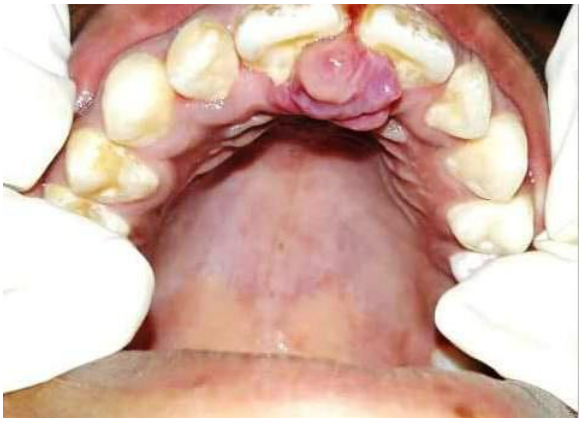

Figure 1a Clinical picture of the lesion

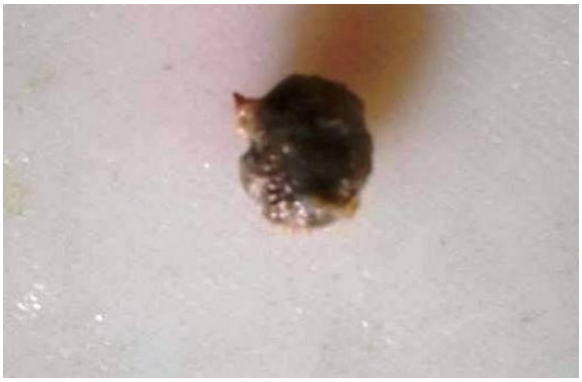

Figure 1b Gross tissue.

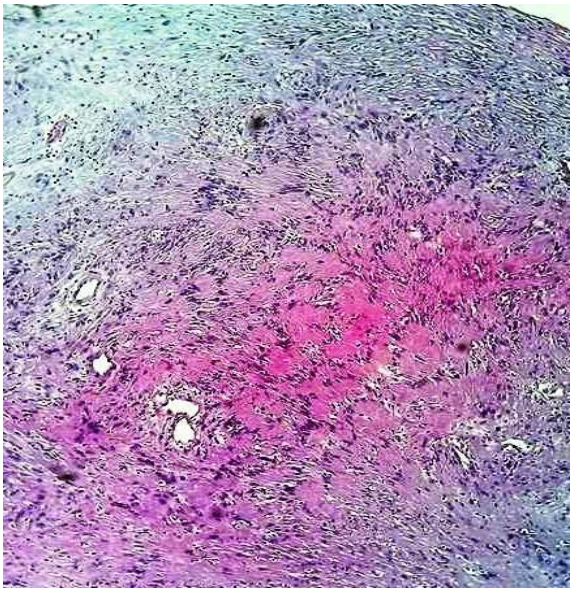

Figure 2a Spidle shaped cells arranged in Antoni A configuration surrounding eosinophillic structures suggestive of verocay bodies (Hematoxylin and eosin staining X10)

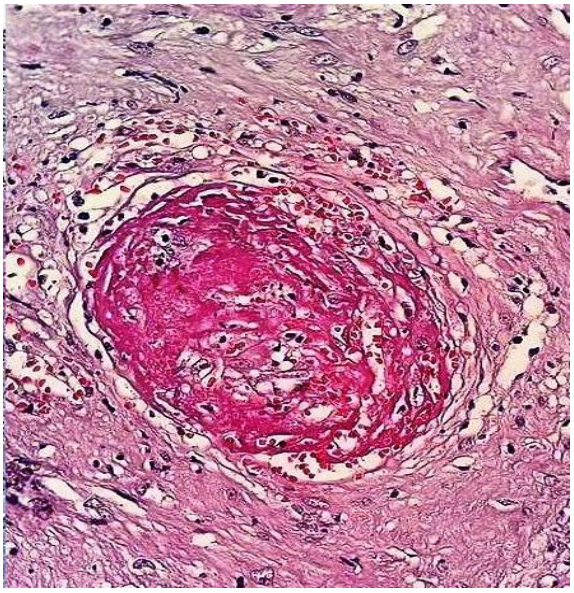

Figure 2b A large thrombus surrounded by atypical spindle cells (Hematoxylin and eosin staining X40)

cases were reported in patients in 4th decade of life except one case in 2013 reported by Muruganandhan in a 22-year-old patient.[4] no case of oral AS has been reported in a pediatric patient so far. This case will probably be the first presentation of oral AS in a pediatric age group patient. The average duration is 10 years; however, few cases showed 2-5 months duration, in present case the duration of 10 months was noted.[3] Oral AS is seen mostly in the tongue followed by buccal mucosa, labial mucosa, palate etc. The present case was reported in the lingual surface of anterior maxillary gingiva.[3,4]

Histopathologically AS shows degenerative features like hemorrhage, hemosiderin pigmentation, cellular atypia, areas of myxoid degeneration.[7] In present case also all the histological features were noted accompanied by Antoni A and B cellular configuration with verocay bodies. Immunohistochemically they are positive for S-100, Leu [7] and myelin basic protein (MBP) confirming their neural origin.[8] Surgical excision is the treatment of choice and no recurrence and malignant transformation have been reported. In present case also 6-month follow-up period was uneventful.

It can be concluded that AS is a rare oral tumor that may be mistaken as malignancy owing to the cellular atypia and other degenerative changes. A proper communication of the surgeon and oral pathologist is mandatory in order to avoid aggressive mode of treatment, furthermore there is a paucity of literature regarding oral AS in a pediatric patient. The aim of this case report is to discuss this unusual tumor in a 10-year-old child.

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