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CASE REPORT

Pediatric schwannoma of the tongue: A case report and review of literature

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Abstract

BACKGROUND

Neurogenic tumors account for about ten percent of all tumors of childhood, and benign tumor originating from Schwann cells is rare in peripheral nerves. Schwannoma of the tongue is quite rare in children.

CASE SUMMARY

We present the case of an 8-year-old male with schwannoma in the anterolateral tongue. The mass was slow-growing for one year with no pain and discomfort. He underwent transoral mass excision under general anesthesia. Gross examination revealed a smooth surfaced, 17 mm × 14 mm × 7 mm sized, encapsulated nodule with a clear resection margin. Schwannoma of the tongue was confirmed by the pathological exam. He reported no motor or sensory change, such as dysgeusia or paresthesia, or phonation difficulty during postoperative 12 mo follow-up.

CONCLUSION

Schwannoma of the tongue is a rare benign neoplasm in childhood. If a painless firm mass is encountered in the tongue of a child, solid tumors like schwannoma should be considered in the differential diagnosis.

Key Words: Schwannoma; Tongue; Child; Pediatric; Neurogenic; Tumor; Case report

Core Tip: Schwannoma of the tongue is a rare benign neoplasm in childhood. If a painless firm mass is encountered in the tongue of a child, solid tumors like schwannoma should be considered in the differential diagnosis. Based on the suspicion of schwannoma, meticulous surgical excision is necessary in terms of functional

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INTRODUCTION

Neurogenic tumors account for 10% of all tumors of childhood[1]. Of these, schwannoma (neurilemmoma), which is a slow growing, benign tumor originating from Schwann cells, is rare in peripheral nerves[2]. About 25% of schwannomas are identified in the head and neck area, and only 1% have intraoral origins, which include tongue, palate, buccal mucosa, lip, and gingiva[3] Although several reports have been issued on this topic of intraoral schwannoma in young patients, no review has been published on pediatric schwannoma of the tongue. Herein, we present a case of pediatric schwannoma of the tongue and review available literature over the last 56 years (from 1964 to 2020). In the English literature over the past 56 years, a total of 17 pediatric cases of schwannoma of the tongue have been reported. Based on a review of these reports, we explored common clinical symptoms, clinical courses, and the differential diagnosis of this disease.

CASE PRESENTATION

Chief complaints

An 8-year-old boy presented at our otorhinolaryngology outpatient clinic with complaints of a slow growing painless mass in his tongue. He had an anterolateral tongue with slow-growing for one year.

History of present illness

He denied all symptoms including pain, dysgeusia, dysphagia, dysphonia, bleeding, and impaired tongue mobility.

History of past illness

The patient had a free previous medical history.

Personal and family history

He denied any family history.

Physical examination

A 15 mm sized submucosal firm, hard, non-tender mass was identified in the right anterolateral side of the tongue (Figure 1A). Overlying mucosa was intact, and cervical lymph nodes were not palpable.

Laboratory examinations

Initial laboratory testing showed no abnormality.

Imaging examinations

No radiological investigations were performed because the mass was easily visible and palpable.

FINAL DIAGNOSIS

Microscopically the tumor was composed of Schwann cells arranged in a cellular palisading pattern (Antoni type A) with Verocay bodies and a second (Antoni type B) looser, disorganized arrangement (Figure 2A). Antoni A areas composed of Verocay

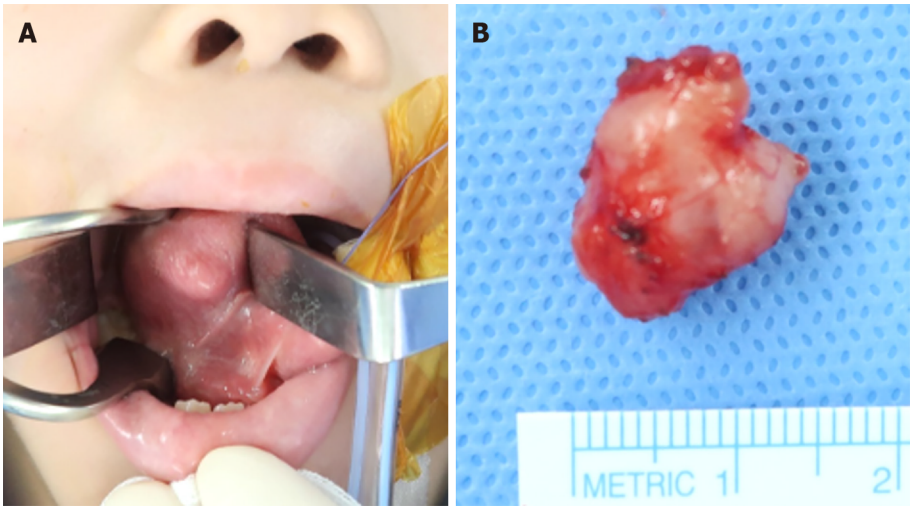

Figure 1 Intraoperative view. A: Preoperative view of the tongue lesion with intact ventral mucosa; B: Macroscopic view of the well-encapsulated, 17 mm × 14 mm × 7 mm sized tumor.

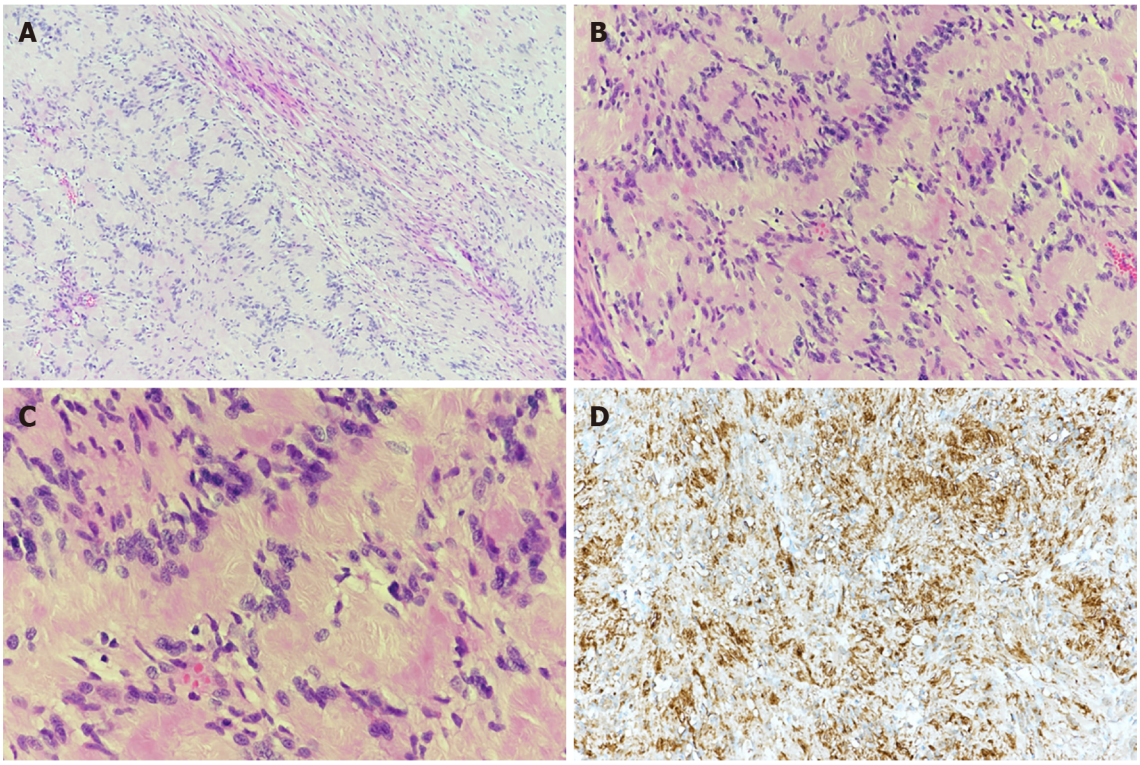

Figure 2 Hematoxylin and eosin stained. A: Histological picture showing the typical biphasic appearance of schwannoma [hematoxylin and eosin (H&E) stained, original magnification × 100]. Densely packed spindle cells (Antoni A areas, left side) with a typical palisading arrangement (Verocay bodies) to loose hypocellular arrangements (Antoni B areas, right side); B and C: Antoni A areas composed of Verocay bodies which consists of a stacked arrangement of two rows of elongated palisading nuclei that alternates with acellular zones (H&E, × 200 and × 400); D: Immunohistochemical staining with S-100 (× 200) was strong and diffusely positive.

bodies which consists of a stacked arrangement of two rows of elongated palisading nuclei that alternates with acellular zones (Figure 2B and C). Diagnosis was confirmed by immunohistochemical staining; tumor tissue was strongly positive for S-100 (Figure 2D).

TREATMENT

He underwent transoral mass excision under general anesthesia. Gross examination

revealed a smooth surfaced, 17 mm × 14 mm × 7 mm sized, encapsulated nodule with a clear resection margin (Figure 1B).

OUTCOME AND FOLLOW-UP

The patient was discharged from the hospital without complications at postoperative two days, and reported no motor or sensory change, such as dysgeusia or paresthesia, or phonation difficulty for postoperative 12 mo.

DISCUSSION

Schwannomas are benign neoplasms arising from any nerve, including autonomic, peripheral, or cranial nerves, but not from optic or olfactory nerves[4]. About 25% of all schwannomas are located in the head and neck, and the parapharyngeal space is the most common site[2,3]. Schwannomas in the oral cavity are uncommon (1%), and usually affect the tongue followed by buccal or vestibular mucosa, soft palate, floor of mouth, gingiva, or lip[5,6]. Tongue schwannomas may occur at any age, but peak incidence is usually seen between 20 and 50 years of age[3]. Reports indicate any part of the tongue may be involved (*e.g.*, ventral, base, or tip)[7-10]. The majority of cases (around two-thirds) involve the anterior, mobile portion, and in the other third, the posterior portion of the tongue base. Among the 21 pediatric cases reviewed, proportions of anterior and posterior locations were almost equal.

During our literature review from 1964 to 2020, we identified 20 cases of pediatric schwannoma of the tongue (Table 1). The 21 cases (including our case) showed no gender predilection [11 males (52.4%) and 10 females (47.6%)]. Age at onset ranged from 7 to 15 years (mean 12 years). The location of tumor was divided into half anterior and posterior. Eleven cases occurred anteriorly and 7 posteriorly; those of other three cases were not mentioned. Schwannoma diameters ranged from 5 to 30 mm and all were resected using a transoral approach. Most patients had no complaints after surgery, though four had symptoms such as snoring, oral bleeding, or a mastication or swallowing difficulty.

Tongue schwannoma can arise from the hypoglossal, lingual, or glossopharyngeal nerves, but it is difficult to determine its origin preoperatively[11]. It has been previously reported most patients are asymptomatic, and that in some the tumor is ulcerative and causes oral bleeding[12]. Typically, if a patient has nerve-related symptoms before or after operation, the origin of the schwannoma can be inferred. However, if a patient is asymptomatic perioperatively, *e.g.*, because the tumor has been growing slowly over several years, the tumor's origin cannot be inferred, and surgical procedure requires meticulous enucleation to minimize nerve injuries. Our patient had a lesion of duration one year, but did not have paresthesia, pain, loss of taste sensation, motor or sensory loss, or phonation difficulties, and postoperatively, did not complain of any complication. Accordingly, we could not determine its neural origin.

Schwannomas are usually solitary, but if a patient has multifocal lesions, (1) Multiple localized neurilemmomas; (2) Neurofibroma in von Recklinghausen's disease; and (3) Schwannomatosis (a non-hereditary disease characterized by multiple subcutaneous and intradermal schwannomas along with variety of intracranial tumors) should be considered[3,13]. The main components of the differential diagnosis in pediatric tongue solid tumor are other benign neoplasms such as hamartoma, choristoma, rhabdomyoma, neurofibroma, lipoblastoma, myoblastoma, and neurilemmoma[14]. In a pediatric tongue lesion series, Sato *et al*[15] reported a high percentage (80%) of vascular and lymphatic lesions and a relatively low percentage of solid tumors (8%)[15]. Unlike adult patients, salivary gland tumor of the tongue is rare in childhood. Therefore, when a pediatric patient is encountered with solid, firm mass in the tongue, we consider solid tumors of neuromuscular origin after excluding lymphovascular lesions. Magnetic resonance imaging can be useful during initial workups in terms of differential diagnosis and determining lesion extents[13]. Characteristically, Schwannoma has a homogeneous well-circumscribed border and does not infiltrate surrounding tissues[4].

Due to the low incidence and nonspecific clinical presentation, the diagnosis of schwannoma is confirmed by histopathologic and immunohistochemical evaluations. Microscopically, the encapsulated tumor typically has a biphasic appearance. Antoni A areas contain Schwann cells densely packed in a palisading pattern with Verocay

Table 1 Pediatric patients and tumor characteristics as determined by review

Ref.	Age (yr)	Sex	Location of tumor	Size (greatest dimension, mm)	Presenting symptoms	Surgical approach
López-Jornet and Bermejo-Fenoll [17], 2005	8	F	Posterior	30	Painless mass	Transoral
Uj[18], 1967	13	F	N/A	N/A	Painless mass	Transoral
Barbosa and Hansen[19], 1984	12	M	N/A	5	Painless mass	Transoral
Akimoto *et al*[20], 1987	14	M	Anterior	10	Painless mass	Transoral
Siar *et al*[21], 1988	13	F	N/A	44	Painless mass	Transoral
Bassichis and McClay[9], 2004	9	M	Posterior	23	Snoring, difficulty breathing	Transoral
Cinar *et al*[10], 2004	7	M	Anterior	10	Painless mass	Transoral
Hsu *et al*[22], 2006	9	M	Anterior	12	Painless mass	Transoral
	12	F	Anterior	16	Painless mass	Transoral
	15	F	Anterior	12	Painless mass	Transoral
Enoz *et al*[23], 2006	7	M	Anterior	25	Painless mass	Transoral
Pereira *et al*[24], 2008	12	M	Posterior	15	Painless mass	Transoral
Karaca *et al*[25], 2010	13	F	Anterior	20	Painless mass	Transoral
Naidu and Sinha[12], 2010	12	M	Anterior	N/A	Oral bleeding	Transoral
Lukšić *et al*[26], 2011	10	M	Posterior	18	Painless mass	Transoral
Husain *et al*[27], 2011	10	F	Posterior	50	Disturbance in mastication	Transoral
Manna *et al*[28], 2012	15	M	Posterior	12	Disturbance in swallowing	Transoral
Bouguila *et al*[29], 2013	15	F	Posterior	28	Oral bleeding	Transoral
Bhola *et al*[30], 2014	14	F	Anterior	15	Painful nodule	Transoral
Moreno-García *et al*[13], 2014	13	F	Anterior	20	Painless mass	Transoral

N/A: No application.

bodies, whereas Antoni type B areas are looser and disorganized. Diagnosis is confirmed by immunohistochemical staining for S-100, SOX10, Leu-7 antigen, vimentin, and glial fibrillary acidic protein[13,16]. In this case, the diagnosis was performed by only S-100 which is a typical marker of Schwann cell.

The treatment of choice for tongue schwannoma is complete surgical excision, which if achieved prevents recurrence[13]. Thus, incomplete excision must be avoided to ensure the preservations of normal speech and swallowing function, especially in children. Malignant transformation is rare[16], and the transoral approach is appropriate for aesthetic restoration[11].

CONCLUSION

Schwannoma of the tongue is a rare benign neoplasm in childhood. If a painless firm mass is encountered in the tongue of a child, solid tumors like schwannoma should be considered in the differential diagnosis. Complete meticulous surgical excision is important in terms of functional preservation and preventing recurrence.

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