Journal of Kerman University of Medical Sciences

JKMU

Journal of Kerman University of Medical Sciences, 2018; 25 (4): 375-381

Schwannoma in the Posterior Hard Palate and Anterior Mandibular Gingiva: a report of two cases

Samira Basir Shabestari, M.D.S. [1], Iman Shirinbak, M.D.S. [2], Shadi Saghafi-khadem, M.D.S. [3], Nosratolah Eshghyar, M.D.S.[4]

1- Assistant Professor, Department of Oral and Maxillofacial Medicine, School of Dentistry, Alborz University of Medical Sciences, Karaj, Iran

2- Assistant Professor, Department of Oral and Maxillofacial Surgery, School of Dentistry, Alborz University of Medical Sciences, Karaj, Iran (Corresponding author; iman.shirinbak@gmail.com)

3- Associate Professor, Department of Oral and Maxillofacial Pathology, School of Dentistry, Mashad University of Medical Sciences, Mashad, Iran

4- Professor, Department of Oral and Maxillofacial Pathology, School of Dentistry, Tehran University of Medical Sciences, Tehran, Iran

Received: 14 December, 2017 Accepted: 12 April, 2018

ARTICLE INFO

Article type:
Case Report

Keywords:
Schwannoma
Neurilemoma
Gingiva
Hard palate

Abstract

Background: Schwannoma is a benign tumor which usually presents as a solitary slow-growing painless mass. Approximately 25–48% of all schwannomas occurs in the head and neck region and is found rarely in oral cavity, about 1% of head and neck cases. Tongue is the most common site, followed by buccal mucosa, palate, floor of the mouth, jaw, gingiva and lips. This tumor may present at any age but it is more prevalent in third and fourth decades of life.

Case Presentation: Herein we report two cases of schwannoma in a 16-year-old woman and an 18-year-old man with asymptomatic pedunculated mass on anterior mandibular gingiva and painful swelling on posterolateral region of hard palate, respectively. The first lesion was discovered during the routine dental visit and the second one was referred by a dentist. They were medically healthy and their dental radiographies showed no particular findings. The provisional diagnosis for the first and second case was benign mesenchymal lesions and salivary gland tumor, respectively. Excisional biopsy was performed for them. Based on histopathological reports, the definitive diagnosis was neurilemmoma. There was no sign of recurrence approximately 2 years following surgical excision of tumors. The purpose of this report is to present two cases of schwannoma in an unusual location for intraoral tumors.

Conclusion: Because of difficulty to diagnose this tumor based on clinical appearance alone, it is suggested that in well-defined peripheral intraoral lesion, schwannoma should be included in differential diagnosis even on palatal or gingival mucosa of young patients. Definitive diagnosis is best stablished by histopathological examination.

Citation: Basir Shabestari S, Shirinbak I, Saghafi-khadem SH, Nosratolah Eshghyar N. Schwannoma in the Posterior Hard Palate and Anterior Mandibular Gingiva: a report of two cases. Journal of Kerman University of Medical Sciences, 2018; 25 (4): 375-381.

Introduction

The Schwannoma, also called as `neurinoma`, `neurilemmoma`, `schwan cell tumor`, or `perineural fibroblastoma`, is a benign, epineurium-encapsulated tumor which develops from the neural sheath's of schwann cells**.** In addition to the typical Schwannoma, other histologically variants of Schwannoma have been described such as plexiform, cellular, epithelioid, and ancient Schwannomas (1). Clinically, it is solitary, smooth-surfaced, with an asymptomatic firm mass, and is usually considered as a slow growing painless nodule (2,3). Approximately 25-48% of all neurinomas are located in the head and neck region, but of these only 1% occur intraorally as an uncommon lesion (1,3)**.** Intraoral neurinoma may present at any age, but it is more frequent between the 3rd and 4th decades of life and there is no sexual predilection accordingly (1,2,4). The most frequent site for the occurrence of this tumor in a decreasing order include tongue, buccal mucosa, floor of the mouth, palatal, vestibular mucosa, lips, jaws, and gingiva (2,4,5). The differential diagnosis includes benign tumor lesions, minor salivary gland tumor (SGT), mucocele, palatal abscess, inflammatory lesions, (epi) dermoid cysts, and granular cell tumor. This lesion is not often observed during clinical practice (6,7). The histological patterns for neurilemmoma are unique and usually appear as an intimate mixture of fusiform spindle cells forming hypercellular Antoni A and myxoid, hypocellular Antoni B areas. Schwann cells are arranged in a palisaded pattern around eosinophilic accelular area (verocay bodie) to form Antoni A. Groups of spindle nuclei and oval-shaped Verocay bodies can frequently be observed (1,3,4). Immunohistochemical (IHC) analysis is useful to verify the neural differentiation of the schwannoma (3). It reveals that all types of schwannomas are positive for the protein S-100, a marker for neural tumors (2,3,4). Schwannomas are usually well-encapsulated, so the treatment of choice is a complete surgical removal of the tumor with preservation of the neighboring structures (1,2,6). The prognosis is good especially in intraoral Schwannoma because recurrences and risk of malignant transformation are rare if tumor is completely excised (3,8).

The aim of this case report is to present two cases of schwannoma in the posterolateral part of the hard palate and at the midline of mandibular gingiva which are uncommon locations for intraoral tumors.

Case Presentation

Case 1

A 16-year-old woman presented to our department for a routine dental visit. She did not have any chief complaints and her medical history was not contributory. On routine examination, there were no significant findings like trauma, bleeding, or discomfort from the lesion. However, intraoral examination revealed an asymptomatic, pedunculated oval mass on lower gingiva at the midline. The lesion was smooth surfaced, and it was covered with normal mucosal color. Its size was approximately 14×3 mm. On palpation, the mass was non-tender, mobile and rubbery in consistency. Teeth in the involved region were vital with positive pulp test. They were free of calculus and caries. The gingiva was pink, non-erythematous and of firm consistency (Figure 1-A). Radiographic examination showed no remarkable findings. From these clinical data, the provisional diagnosis was

established as reactive lesions (Peripheral Ossifying Fibroma) or Benign Mesencymal Tumors (BMT). After obtaining written informed consent, an excisional biopsy was done under local anesthesia using lidocaine hydrochloride 2% with adrenaline 1:100.000. Grossly, the lesion was an encapsulated rubbery mass with mucosal lining. After tissue processing, microscopic evaluation revealed that an oral mucous surface was covered with stratified squamous, parakeratinized epithelium and retepeges. In the connective tissue, a benign tumor with lobular pattern was composed of elongated spindle and wavy cells. The tumor was circumscribed by a thin well-defined fibrous connective tissue capsule. In the background tissue, we observed some areas with Antoni type A aspect, forming Verocay bodies (Figure 2-A). The IHC examination for S-100 protein was done and showed positivity in the tumor cells in a diffuse pattern (Figure 3-A). Based on the clinical and histophatlogical view, the final diagnosis was neurilemmoma. The therapeutic modality was a complete surgical removal. The patient underwent follow-up visits. The postoperative recovery was uneventful. There were no signs of recurrence approximately 1 year and 8 months following surgery.

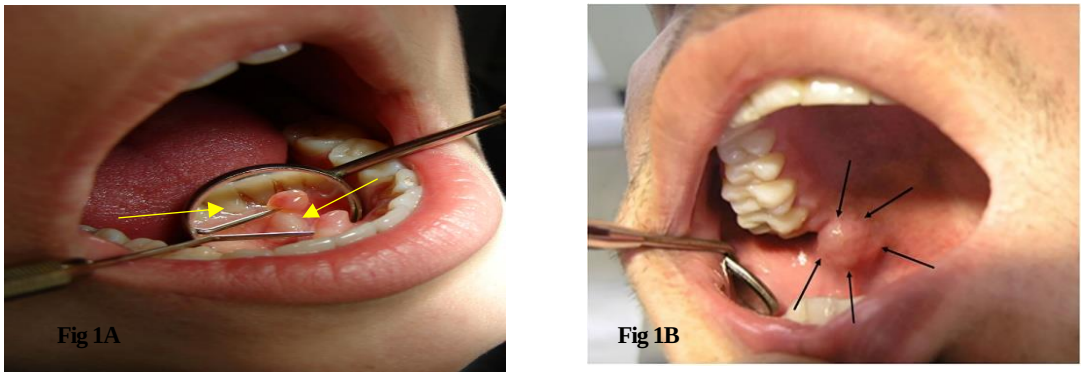

Figure 1. Intraoral view of cases: A) Mirror view of the lesion as pedunculated mass with smooth surface that is located in the midline of mandibular gingiva. B) The encapsulated mass observed in the right side of posterior aspect of the palate. The borders are marked.

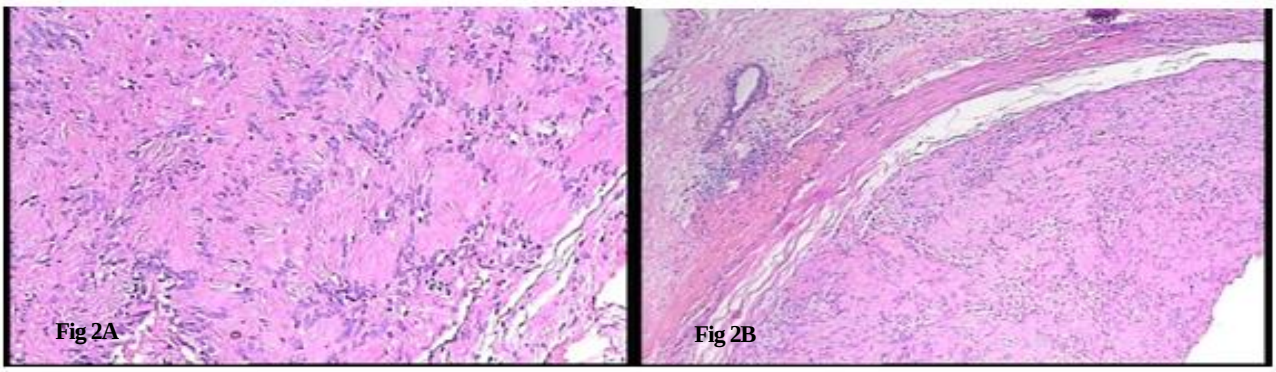

Figure 2. Histopathological images of Schwannoma. A) Case 1: The Schwann cells of the Antoni A tissue form a palisaded arrangement around acellular zones known as Verocay bodies (H&E, ×100). B) Case 2: Low-power view showing encapsulated tumor with well-organized Antoni A. (H&E, ×40)

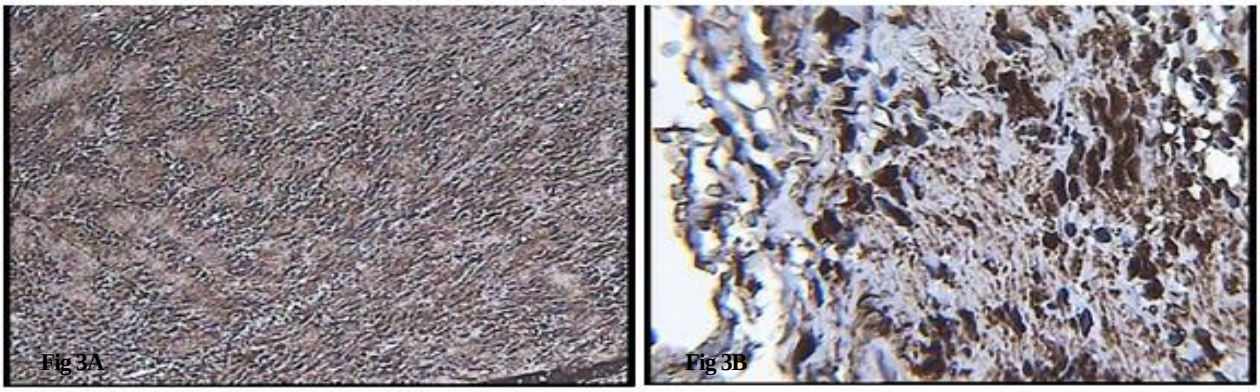

Figure 3. IHC analysis of Schwannoma : A) Case 1: Low-power view showing positive Immunohistochemical stain for S-100 (×100), B) Case 2: Demonstrating spindle-shaped cells that are positive for S-100 protein (×400)

Case 2

An 18-year-old man referred to our department with the chief complaint of a painful swelling of the palate that had been present for 6 months. He took antibiotic and analgesics during 1 month before his visit. The lesion was persistent and firm without an increase or decrease in size over a period of 7 months. There was no history of trauma to palatal region. There was no report of hemorrhage or discomfort from the lesion. Familial history and review of systems were non-contributory. On extraoral examination, there were no significant findings and also no lymphadenopathy was observed. Intraorally, the lesion was a solitary round-shaped mass seen on the right side of posterolatral portion of the hard palate. Mucosa over the swelling was similar in color and texture with the surrounding mucosa. Its size was 20 x 10 mm with nodular swelling. The mass was well-defined and smooth-surfaced. On palpation, the nodular mass was encapsulated, slightly tender, firm-to-rubbery in consistency with no evidence of any discharge. There was neither a carious tooth nor any fistula (Figure 1-B). The adjacent teeth responded positively to pulp testing. Dental radiographies showed no particular findings. Owing to the clinical and paraclinical dates, a provisional diagnosis of SGT or BMT was made. After obtaining written informed consent, the patient underwent excisional biopsy of the mass under local anesthesia (lidocaine hydrochloride 2% with adrenaline 1:100.000). After complete excision of the lesion, the gross specimen underwent tissue processing. Histological view showed benign neoplastic proliferation of spindle-shaped Schwan cells with Antoni A and Antoni B pattern and eosinophilic region of verocay bodies. A thick fibrotic capsule surrounded the lesion (Figure 2-B). The IHC analysis was diffusely positive for protein S-100 (Figure 3-B). With all these findings, a final diagnosis of schwannoma was confirmed. The therapeutic intervention was surgical excision. The patient underwent clinical control. The postoperative recovery was uneventful. Thereafter, no sign of recurrence was seen 2 years and 1 month following surgery.

Discussion

Schwannoma is an infrequent, slow-growing, and is usually presented a solitary well defined mass (1,2). The most frequent site of occurrence for intra-oral schwannoma is the tongue which is usually manifested as a submucosal nodule (4,6).

In the present cases, the lesions were single, rubbery in consistency and were covered with normal mucosa. These cases did not have any particular features to distinguish them from other benign soft tissue masses which occur in young

female and male patients. Their histopathologic findings were typical for schwannoma and in these cases there were not any degenerative changes or cellular atypia (9).

In a study conducted by Salehinejad, a total of 37 case reports concerning oral schwannoma were reviewed. The majority of case reports in this study focused on one case report regarding oral schwannoma. Interestingly our article reported two cases (10).

The schwannoma of oral cavity is usually present in soft tissues such as tongue followed by buccal mucosa and anterior portion of the palate (4). A study by sakanashi during 2000 to 2011 reported four cases of intra oral Schwannoma which were observed in the gingival mucosa (9). Wakoh M et al reviewed 22 cases of schwannomas. The frequency in palate and gingiva region was 7 and 1 respectively (11). The occurrence of this tumor in the posterolateral aspect of the hard palate and at the midline mandibular gingiva, as our study, is still uncommon.

Gender distribution of oral schwannoma in various studies is different. Some studies such as the study by Salehinejad J et al (2016) and Martins MD (2009) found no significant correlation between the occurrence of oral schwannoma and gender (2,10). Our study reported two cases in both genders. However, some studies such as the study by Kumar D (2012) and Rahpeyma A et al (2012) showed a slight tendency toward women (6,12).

Based on various researches, the age of patients ranged from 3 to 70 years. Oral schwannomas may present at any age and the peak of incidence is usually seen in the 25 to 45-year-old groups (1,2,4,6,12). So, we assume that our cases may be of interest regarding age.

Presentation of head and neck schwannoma depends on the site of the tumor and its effect on nerve. Generally, symptoms are due to local mass effect or the dysfunction of the nerve they arise from. Patients may experience pain, neuropathy of cranial nerves, dysphagia and abnormal voice changes (2,12). Clinically, the most intraoral Schwannomas are asymptomatic and usually encountered in routine intraoral examination like our first patient (4). In a review study done by Salehinejad J et al (2016), only 3 cases of painful Neurilemoma were observed in the 37 studied articles. According to several cases, palatal swelling may cause pain and discomfort (10). A study reported that due to pressure effect of the mass on an adjacent nerve, patient may experience paresthesia (12). In our study, the first case was asymptomatic, but the other case had pain in palatal region. Therefore, clinicians must be aware of various signs and symptoms of oral Neurilemoma.

As the final clinical diagnosis of oral schwannoma is difficult, it seems that identification of the tumor is surprising (10). In case 1, which was accidentally discovered during dental examination, differential diagnosis was made with reactive lesions (such as Peripheral Ossifying Fibroma, epulis) and BMT. As mentioned previously, a probable diagnosis for case 2 was SGT. Pleomorphic adenoma is the most prevalent benign salivary gland neoplasm in palatal region. It is usually presented as a non-tender, slow-growing swelling with a smooth surface that can be ulcerated due to trauma (1,3). Mucoepidermoid carcinoma is the prevalent malignant SGT. Minor salivary glands especially in hard palate constitute the second most common site for this carcinoma after parotid glands. Clinically, mucoepidermoid carcinoma can manifest itself as an asymptomatic swelling (3,6,7). Palatal abscess was ruled out at first because of positive vitality test for the right side maxillary teeth and lack of response to a course of antibiotic therapy. There was not any history of previous trauma to this region.

At first, no specific hypothesis concerning intraoral schwannoma was formulated because of its uncommonness, especially at the posterolateral part of hard palate and lower gingiva and nonspecific clinical presentation. Achieving an accurate preoperative diagnosis was reasonably difficult, so biopsy was done (2,13). The final diagnosis was made with the aid of microscopic view and immunostaining analysis.

Schwannoma in histopathologic view consists of two basic tissue types. Antoni type A presents as fascicles of palisaded spindle-shaped Schwann cells streaming around Verocay bodies and the second tissue type, Antoni type B, lacks this pattern and includes less cellular and more randomly arranged cells in a loose myxomatous stroma (14). Histologically, our case report was similar to the study by Rajendran in which the tumoural tissue consists of the so-called Antoni A and B type pattern with more myxoid consistency (15). In our cases, Antoni A was more prominent than Antoni B and two cases were circumscribed by a thin well-defined fibrous connective tissue capsule like cases presented by kumar et al, karatas et al, and Moradzadeh khiavi et al (6,16,17). In a case report by Yaga similar to our cases, there was an encapsulated mass which showed a connective tissue stroma composed of spindle cells with palisaded pattern around eosinophilic areas (18). In our study similar to reports by Purwaw et al, Chandra et al and Eroglu et al (19-21) and in contrast to Nishijima Sakanashi et al (9), mitotic activity was not observed. In contrast, Upadhay reported a case with some areas of cystic degeneration and hemorrhage (22). Immunostaining analysis was done and our two cases were positive for S-100 protein (16,17).

Excisional biopsy is indicated in solitary slow growing lesions. As schwannomas do not infiltrate the parent nerve, they can usually be separated from it (1,9,13). So, the treatment of choice for this tumor is surgical excision (2-4). In our cases, excisional biopsy was done for diagnostic and therapeutic purposes. The biopsied lesions were well encapsulated and pedunculated, so they could be totally excised. With complete excision of the tumor, there is a low risk of recurrence. Based on available literature, recurrence was seen in none of the cases similar to our study (1,2,10,13).

Conclusion

We reported two rare cases of schwannoma located in the posterior hard palate and anterior mandibular gingiva. As the clinical diagnosis is difficult, there is a need for histopathological analysis. This lesion must be differentiated from other benign lesions that appear in the same regions. We should bear in mind that a slow-growing, well circumscribed mass of oral cavity even in the palatal mucosa may be a Schwannoma.

Acknowledgment

The authors would like to thank the contribution of Oral Medicine Departments of Tehran and Mashhad Dental Schools.

References

1. Bayindir T, Kalcioglu MT, Kizilay A, Karadag N, Akarcay M. Ancient schwannoma of the parotid gland: a case report and review of the literature. J Craniomaxillofac Surg 2006; 34(1):38-42.
2. Martins MD, Anunciato de Jesus L, Fernandes KP, Bussadori SK, Taghloubi SA, Martins MA. Intra-oral schwannoma: case report and literature review. Indian J Dent Res 2009; 20(1):121-5.

3. Neville BW, Damm DD, Allen CM, Bouquot JE, Chi AC. Oral and Maxillofacial Pathology. 4th ed. Missouri: St Louis: Elsevier Health Sciences; 2016.
4. Lopez-Carriches C, Baca-Perez-Bryan R, Montalvo-Montero S. Schwannoma located in the palate: clinical case and literature review. Med Oral Patol Oral Cir Bucal 2009; 14(9):e465-8.
5. Sharma K, Sharma P, Maheshwari P, Gupta A, Bansal P. Neurilemmoma of lower gingival mucosa: a rare case report. Indian Journal of Contemporary Dentistry 2016; 4(2):48-51.
6. Kumar P, Meghashri K. Schwannoma of the hard palate: a case report and review of literature. Journal of Advanced Oral Research 2012; 3(1):23-8.
7. Shabestari SB, Shirinbak I, Agha-Hosseini F. Maxillary metastasis of a medullary thyroid carcinoma in a 21-year-old woman 7 years after thyroidectomy. J Oral Maxillofac Surg 2012; 70(6):1495-9.
8. Schackert HK, Agha-Hosseini F, Gorgens H, Jatzwauk M, von Kannen S, Noack B, et al. Complete homozygous deletion of CTSC in an Iranian family with Papillon-Lefevre syndrome. Int J Dermatol 2014; 53(7):885-7.
9. Nishijima Sakanashi E, Sonobe J, Chin M, Bessho K. Schwannoma located in the upper gingival mucosa: case report and literature review. J Maxillofac Oral Surg 2015; 14(Suppl 1):222-5.
10. Salehinejad J, Sahebnasagh Z, Zare-Mahmoodabadi R, Sahebnasagh Z, Ebrahimi S. Oral schwannoma: a systematic review of the case reports. Journal of Dental Materials and Techniques 2016; 5(2):99-111.
11. Wakoh M, Yonezu H, Otonari T, Sano T, Matsuzaka K, Inoue T, et al. Two cases of schwannoma with marked cystic changes. Dentomaxillofac Radiol 2005; 34(1):44-50.
12. Rahpeyma A, Jafarian AH, Khajeh Ahmadi S, Sarabadani J. A schwannoma of the soft palate in a child: histological and immunohistochemical features and surgical method. Iran J Otorhinolaryngol 2012; 24(67):95-9.
13. Shabestari SB, Shirinbak I, Azadarmaki R. A Comprehensive Look at Oromaxillofacial and Laryngopharyngeal Cancers. In: Mehdipour P, editor. Cancer Genetics and Psychotherapy. Cham: Springer International Publishing; 2017. p. 531-87.
14. Gnepp DR. Diagnostic Surgical Pathology of the Head and Neck. 2th ed. Philadelphia: Saunders Elsevier; 2009.
15. Rajendran R. Benign and malignant tumors of the oral cavity. Shafer's Textbook of Oral Pathology. 7th ed. Elsevier India; 2009.
16. Moradzadeh Khiavi M, Taghavi Zenouz A, Mesgarzadeh AH, Sabetmehr O, Mahmoudi SM, Kouhsoltani M. Schwannoma in the midline of hard palate: a case report and review of literature. J Dent Res Dent Clin Dent Prospect 2014; 8(2):114-7.
17. Karataş M, Sarıkaya Y, Doğan S, Erdoğdu İH, Bulut HT, Yücetaş SC. A slow growing mass of hard palate: schwannoma-case report. J Clin Med Case Reports 2015; 2(1): 1-4.
18. Yaga US, Mengji AK, Besta R, Gollamudi N. Rare case of palatal schwannoma: case report and review of literature. J Indian Acad Oral Med Radiol 2015; 27:484-7.
19. Purwaw P, Dixit J, Singhal D, Bhartiya K, Vashistha M, Singh M. Schwannoma of gingiva: a rare case report. J Orofac Res 2014; 4(3):189-92.
20. Chandra M, Singh P, Venkatchalam VP. Tongue schwannoma: a case report with review of literature. JK- Practitioner 2013; 18(1- 2):28-34.
21. Eroglu CN, Tun SK, Gunhan O. Soft tissue schwannomas of the hard palate and the mandibular mentum. Case Reports in Dentistry 2017; 2017: 1-4.
22. Upadhyay S, Bhavthankar J, Mandale M, Humbe J. A diagnosis of an unusual lower lip swelling: Schwannoma. Niger Postgrad Med J 2017; 24:191-3.