

Schwannoma in the maxillary sinus and buccal space: Case report

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Abstract (J. Kor. Oral Maxillofac. Surg. 2009;35:494-498)

Schwannomas are tumors which originate from the neuroectodermal Schwann cell of cranial, intraspinal, peripheral and autonomic nerve sheaths, and they are solitary, benign, slow growing and well encapsulated neoplasm. Schwannomas are usually asymptomatic. No strong gender or age predominance exists.

The incidence of extracranial schwannomas in the head and the neck region varies from 25~45%. In addition, schwannomas are rare in the maxillary sinus or buccal space.

In this paper, it diagnosed and treated a 54-years old female patient, who had schwannoma in the maxillary sinus derived from infraorbital nerves, the branch of the left trigeminal nerve, and a 19-years old male patient, who had schwannoma arose in the buccal space derived from the buccal branch of the right facial nerve. There was no particular complication except sensory extinction of the nerve in the female patient and paralysis by the nerve in the male patient. It is determined those two cases of schwannoma in the rare portion is valuable and herein, it reports those with literature discussions.

Key words: Schwannoma, infraorbital nerve, facial nerve

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Introduction

A schwannoma arises from the neural sheath of Schwann cell in cranial, peripheral or autonomic nerves but not in olfactory and ophthalmic nerves, which have no Schwann cell. It is a rare benign tumor, well-wrapped in capsule which gradually grows. This schwannoma, also known as a neurilemmoma, is about 6~8% of intracranial tumors and is rarely transformed into malignant tumor. It can arise from all parts of the body, however, 25~45% of intracranial schwannoma arises in the head and neck area such as oral cavity, mastoid, middle ear, larynx and paranasal sinuses¹⁾.

There is no particular age or sex in the onset of this disease and most patients have no symptom. Some patients complain of pain or paresthesia when there is central growth in the external wall of the nerve sheath compressing adjacent nerves²⁾. Occasionally, there are symptoms including hearing loss, nasal atresia, dysphagia, hoarseness, headache and tinnitus along with nerves related secondarily, however, it is nonspecific as a

symptom for preoperative diagnosis of a neurologic lesion. FNA cytology, CT scan and MRI image might be restrictive but helpful to diagnosis of schwannoma, however, final diagnosis could be made with postoperative histological findings.

Intracranial schwannoma mostly arises from olfactory nerves, followed by trigeminal nerves and it is rare in facial nerves.

Authors experienced a schwannoma within the maxillary sinus originated from infraorbital nerve, the branch of the trigeminal nerve, and that arising in the buccal space originated from buccal branch in the facial nerve. It is determined those two cases of schwannoma in the rare portion is valuable and herein, it is about to report those with literature discussions.

Case Report 1

The patient is a 54-years old female with a chief complaint of swelling in the left maxillary gingiva since 2 months ago. In the first visit, she had a history of dental extraction of #26 and #27 teeth 2 weeks ago and had no pain or discomfort. She had swelling in the left maxillary facial portion and it observed swelling in the left buccal mucosa in the oral cavity. There was no preoperative hypoesthesia in the left face.

It took the with-enhancement facial CT and observed about 6 cm mass, had a relatively good boundary with low density in

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the left masticator space. By that mass, the left maxillary sinus was pushed forward and masseter muscle, pterygoid muscle and lower jaw were pushed backward. In addition, the mass was located up to the infraorbital fissure. There was no calcification or ossification within the mass and on the postcontrast enhanced CT scan, that mass showed mild enhancement and there was no destruction of adjacent bones. And then, it was not observed any enlargement of lymph node (Fig. 1).

In outpatient treatment, it performed an incisional biopsy for whitish gray soft tissue in the left maxillary sinus portion and she was diagnosed with chronic inflammation with fibrosis without particularity.

It carried out an operation under general anesthesia. It used an intraoral vestibular incision & Caldwell-Luc approach to minimize postoperative scar after resecting the mass, and used a subciliary approach in order to confirm the status of destruction in the left orbital base. It checked and removed bone chips regarded as the maxillary sinus wall. It could identify the mass strongly connected with infraorbital nerves, the maxillary nerve branches of trigeminal nerve, by approaching the inside

of the maxillary sinus. It was almost impossible to separate the mass and infraorbital nerves so it removed the mass together with the part of nerves. It excised the brownish, focally myxoid and soft mass (7.5 × 4.3 × 2.5 cm) and the mass had no necrotic lesion. It was diagnosed as a schwannoma by the frozen section.

It was found that the orbital base was slightly expanded toward the eyeball and there was no status of bone destruction in the base. Therefore, it was unnecessary to reconstruct the orbital base due to this operation.

At histological finding, to some extent, it presented a typical manifestation of schwannoma such as palisaded arrangement of Schwann cell and Antoni A and B area in H&E staining, however, it was confirmed as schwannoma through S-100 protein positive reaction and SMA negative reaction (Fig. 2).

After the operation, there has been no particular symptom except sensory extinction in the left maxillary sinus. After 1 year excision of mass, the alveolar bone of left maxilla was regenerated. The patient received dental implant treatment (Fig. 3).

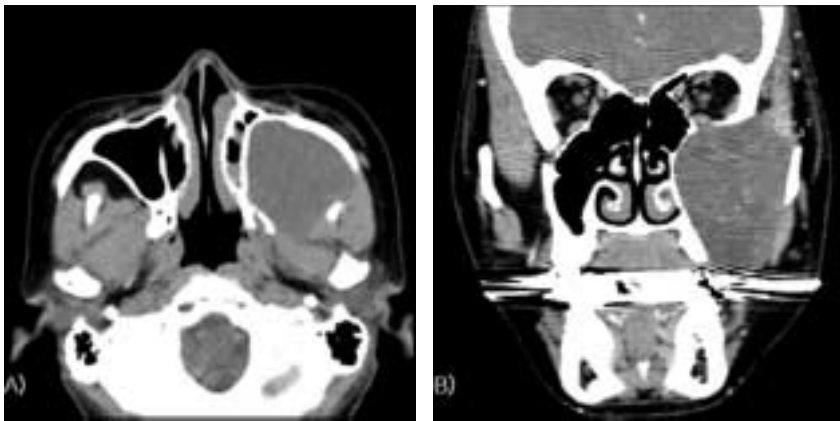


Fig. 1. Axial(A) and coronal(B) CT of patient showing a well-circumscribed large mass measured approximately 7×4×2.5cm invading left maxillary sinus, floor of orbit and infratemporal fossa.

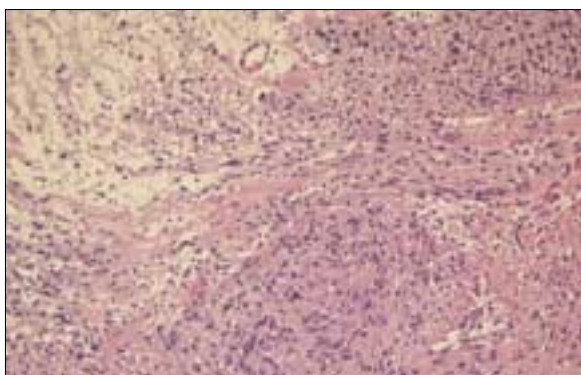


Fig. 2. Microscopic findings. High-power view shows schwannoma architecture consisting of spindle-type cells in a diffuse Antoni A pattern, with a few areas of Antoni B pattern(H&E stain, ×200).



Fig. 3. Panoramic view of 1st dental implant installation, after 1 year excision of mass.

Case Report 2

A 19-years-old male patient visited the dental clinic for evaluation of swelling on his right cheek. He said that he recognized it about 2~3 years ago. Then, in palpation, it was found a hard indurated, painless, slightly movable mass with nodular form and he reported no pain or discomfort caused by that mass. In his physical examination, he had no facial nerve paralysis or hearing loss.

It performed the FNAC for the mass on his right cheek and there was no evidence of a malignant cell.

It took CT and MRI and observed a well defined oval solid mass with $2 \times 2 \times 4$ cm on the right buccal space on MRI result. The mass showed hyperintense signal than surround muscles in T2 weighted images and isointense signal compared with surround muscles in T1 weighted images. The right parotid gland was transposed to the upper side and there was no evidence of cystic change and hematoma. There was a high possibility of lesser salivary gland-derived benign tumor and also of Schwannoma (Fig. 4).

It carried out an operation under general anesthesia. To secure the visual field for this operation, it used a modified blare incision and checked facial nerves using a nerve stimulator. Then, it confirmed the mass under the right parotid gland surrounding the buccal branch of facial nerves by blunt dissection. It was inevitable to remove the mass together with the part of the buccal branch for those could not be separated. It resected rubbery nodule, grayish solid mass ($6.0 \times 2.2 \times 2.1$ cm) wrapped by capsule of bulky and yellowish gray section. The nerves removed were not reconstructed.

On the result of histological findings, it was firmly diagnosed as a typical schwannoma by H&E staining (Fig. 5).

After the operation, there was a partial paralysis on the right lobule of nose and it conducted postoperative rehabilitation.

Discussion

The schwannoma, firstly introduced by Verocay in 1908, is a benign tumor neurologically originated from Schwann cells wrapped the nervous tissue and slowly grow³.

In the result of H&E staining, schwannoma is characterized by the pattern of Antoni A and B area and Verocay body. The typical cell shape of schwannoma is fusiform with a long nucleus. Antoni A area is composed of fusiform cells with parallel rows of a palisade arrangement of the nucleus. Antoni B area has an oviform cell with loose arrangement between mucous substrates and has an intracellular space structure vacuolized. Occasionally, it causes myxoid change accompanied by vitrification around vessels in the prolonged tumor, called an ancient schwannoma.

In addition, a schwann cell-derived tumor is mostly positive in S-100 immunohistochemical staining and S-100 protein positive reaction is a reliable histological feature in the diagnosis of schwannoma⁴.

Due to nonspecific signs and symptoms of schwannoma, it is difficult to diagnose it by laboratory tests. Therefore, it has been introduced FNA cytological technique, MRI and CT image as a diagnostic method for schwannoma. It has been reported that the diagnosis using FNAC characterized by the presence of spindle cells has only 17.6% of accuracy, not so high⁵. To increase the accuracy of diagnosis using FNAC, it requires a high quality of specimen and is important to have an experience of a cytopathologist⁵. Therefore, it is recommended that FNAC is used for excluding the possibility of



Fig. 4. T2-weighted magnetic resonance image revealing $2 \times 2 \times 4$ cm mass involving right side buccal space. Mass is enhanced homogeneously by high signal intensities.

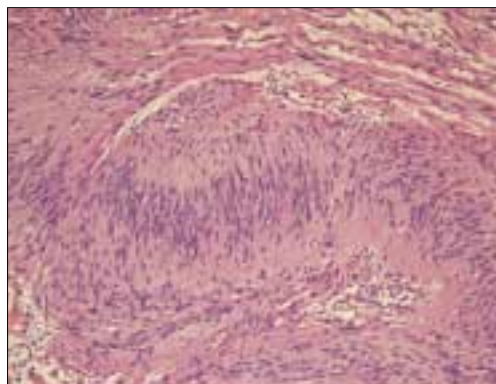


Fig. 5. Photomicrograph of schwannoma showing well-organized Antoni A tissue with adjacent myxoid and less organized Antoni B tissue(H&E stain, $\times 200$).

malignancy rather for the exact diagnosis of schwannoma. Through the FNA conducted in the case 2, it confirmed that there was no malignant cell; however, it had no efficacy on the diagnosis of schwannoma. On CT image, schwannoma was well-wrapped and had a relatively clear fusiform shape. The characteristics of MRI image of schwannoma include the pattern of isointense T1 signal and increased and slightly heterogeneous T2 signal. Besides, after injection of gadolinium, the mass showed better contrast⁶. In the case 1, schwannoma was suspected through CT results. The signal pattern of schwannoma was determined by MRI image in the case 2 and it could diagnose that as a schwannoma. When the diameter of the nerve derived is big, the ultrasound is a very useful diagnostic tool. It has been reported that it could be used for identifying that the mass is often connected with the nerve related, however, its diagnostic rate by ultrasound is low⁴.

Neoplasms originated from cells composed of the nerve sheath are schwannoma and neurofibromatosis. Schwannoma typically arises solely and has a well-wrapped shape. Its transformation to malignant cell is very rare and its recurrence was low if it is completely removed. Neurofibromatosis is commonly multicentric and only 4% of it is wrapped by capsule. Compared to schwannoma, it is more difficult to separate nerves and the mass and has high rate of transformation to malignant cell. It is often related to Von Recklinghausen's disease, and in this case, it shows about 8% of malignant transformation⁷. Schwannoma derived from infraorbital nerves arises within the orbit or maxillary sinus. Intraorbital schwannoma, 1~4% of orbital tumor within the orbit, is generally developed in supraorbital or supratrochlear nerves, and is rare in infraorbital nerves⁸. It is the mass arose from sensory nerves so it does not mostly affect eye movement or visual acuity if it arises within the orbital. The main symptoms are exophthalmos and pain. The mass slowly grows and does not invade surrounding bone structure; however, it sometimes pushes or corrodes the bone wall by pressure.

It is rare to find schwannoma in the paranasal sinus, especially in the maxillary sinus⁹. The symptoms of schwannoma in the maxillary sinus are cheek swelling, nasal atresia, downward transversion of the palate and pain, rarely accompanying exophthalmosis. For the mass slowly grows, it mostly causes the expansion of the maxillary sinus when the size is bigger, and most bony margin is maintained⁷. For the treatment, it should completely resect the mass. If schwannoma compresses the orbital base to elevate the orbital structure and to destroy the orbital base, orbitotomy might be required.

The case 1 presented infraorbital nerve-derived schwannoma in the left maxillary sinus. It completely resect the mass and

there was no complication except paresthesia. The orbital base was elevated to the eyeball by compression; however, there was no destruction of bone. Then, it was unnecessary to reconstruct the orbital base. In addition, there was no limitation for eye movement and no abnormal visual acuity.

Within the buccal space, diverse structures exist such as salivary gland, blood vessel, lymph glands, connective tissues, muscle and nervous tissue. It has been reported various lesions originated from those structures. Neurofibroma has been often reported as a nervous tissue-derived mass; however, it is very rare to find schwannoma within the buccal space.

It has been known that the facial schwannoma arises along with the route of facial nerves, the clinical symptoms of it within the temporal bone, the majority of facial schwannoma, are facial nerve paralysis and hearing loss, and the main clinical symptoms of facial schwannoma outside the temporal bone is a lump of parotid gland¹⁰. It has been also known that facial nerve schwannoma is less than 5% of the cause of facial nerve paralysis and 39% of the schwannoma patient presented facial nerve paralysis.

The patient in the case 2 is a very rare instance of schwannoma arose in the buccal space derived from the buccal branches of facial nerves. Before the operation, he had no facial nerve paralysis due to nerve compression by central growth in the external wall of the nerve sheath and it removed a part of the buccal branches during the operation. Then, there was a postoperative right alinasal paralysis. It could consider immediate reconstruction or postoperative rehabilitation for nerve sacrifice in the incision process of facial schwannoma. In this case, it conducted rehabilitation and followed up the progress, therefore, it confirmed that the activity of dilator naris muscles, which was decreased by paralysis immediately after the operation, was improved.

There are various operational approaches depending on the location or size of the mass, personal preference of a dentist and related nerves. In our clinic, it used modified blare incision in the case 2 so it secured a good visual field and aesthetic results.

It is still open to discussion in some aspects for the treatment of facial schwannoma. Several researchers persisted conservative treatment until there is a precise evidence for facial palsy if it is little or none or if the mass slowly grows with no symptoms. Also, there was a report that, in the case of complete incision for the aged or the patient unendurable to long operation, although it needed so long period for follow up, to treat facial schwannoma in the intratemporal bone by subtotal or neat total resection is proper to manage it without malignant transformation³.

However, the ideal treatment of radioresistant schwannoma is to prevent reoccurrence and to resect it completely while conserving the nerve related in order to minimize nerve dysfunction. In addition, total resection with parotidectomy is the most precise treatment when facial schwannoma rarely arises in the parotid gland⁵⁾.

In the case of facial schwannoma, it has been reported the easy separation between the mass and nerves related only in several cases. Besides, there are some methods for reconstruction of facial nerves such as greater auricular n. interpositioning graft, sural nerve interposition graft and hypoglossal-facial nerve anastomosis if facial nerve is resected¹⁰⁾. However, there is the case of postoperative facial nerve dysfunction despite of efforts to conserve or to reconstruct it with the nerve related.

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