Case Report

Evaluation of cystic schwannoma of masticator space – A case report

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Abstract

Schwannoma is a benign neoplasm originating from the neural sheath and occurring in up to 45% of the cases of extracranial neurogenic tumors. Schwannomas of masticator spaces are rare. This article reports the case of an intraoral schwannoma occupying the masseteric and buccal spaces of a 55-year-old woman, for whom detailed imaging investigations were performed, comprising conventional extraoral radiographs, ultrasound, and plain and contrast enhanced magnetic resonance imaging. The lesion was surgically resected.

Keywords: Conventional extraoral radiographs, magnetic resonance imaging, masseteric and buccal spaces, schwannoma, ultrasound

Introduction

The term "schwannoma," coined by Harkin and Reed in 1968, alludes to any benign, circumscribed, encapsulated perineural tumor of the peripheral nervous system. First described by Jose Juan Verocay, an Uruguayan physician in 1910 as "neurinoma," this tumor also goes by the name of neurilemmoma, neurolemmoma, schwanoglioma, peripheral glioma, and peripheral nerve sheath tumor.[1]

Case Report

A 55-year-old-woman reported to the Department of Oral Medicine and Radiology with a swelling on the left side of face since 6 months. Her medical history was noncontributory. Dental history revealed multiple uneventful visits for extraction of teeth 5 years ago. The swelling had started after consuming a nonvegetarian meal. There was no history of pus discharge, pain, numbness, or other associated symptoms. Extraorally [Figure 1a], it extended from the malar region up to the inferior border of the mandible on the left side, measuring approximately 3 cm × 4 cm in dimension and causing gross facial asymmetry with considerable trismus. Intraorally [Figure 1b], there was an irregular exophytic growth present in the left retromolar trigone region distal to 26, causing vestibular obliteration, and a diffuse swelling in the adjoining posterior buccal mucosa. It was firm in consistency, nontender, and nonfluctuant on palpation. Fine-needle aspiration cytology (FNAC) yielded a brownish straw-colored blood-tinged fluid. Initial radiographic investigations advised were conventional extraoral film radiographs (due to limited mouth opening), and later ultrasound and magnetic resonance imaging (MRI) with and without contrast were performed. Orthopantomogram revealed extensive destruction of the alveolar process of maxilla distal to 26 and anterior border of the ramus of the mandible [Figure 2a]. Paranasal sinus view showed complete opacification of the left maxillary sinus with destruction of its posterolateral wall [Figure 2b]. Ultrasonography [Figure 3] revealed a thick-walled soft tissue mass with central

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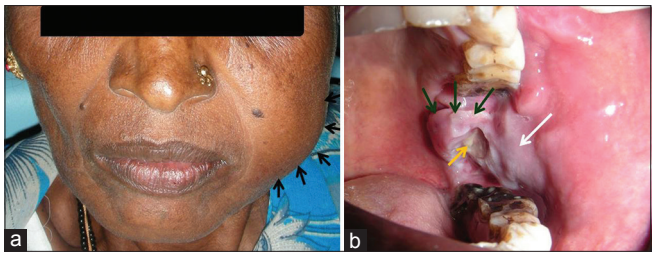

Figure 1: Extraorally (a), an ill-defined swelling extending from the malar region upto the inferior border of the mandible on the left side (short black arrows) was seen. Intra-orally (b), an irregular exophytic growth (green arrows) was present in the left retromolar trigone region distal to 26, causing vestibular obliteration, and a diffuse swelling (white arrow) in the adjoining posterior buccal mucosa. The deepest part of the growth opposite 38 was laden with food deposit (yellow arrow)

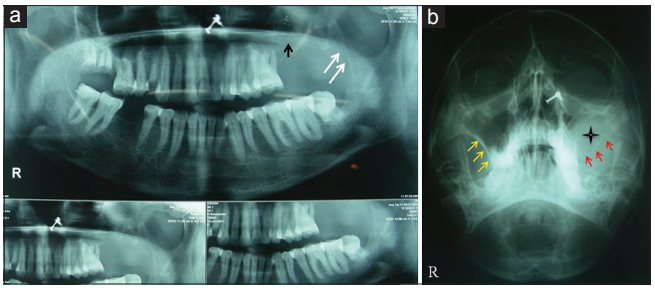

Figure 2: OPG revealed extensive destruction of the alveolar process of maxilla distal to 26 (black arrow) and anterior border of the ramus (white arrows) of the mandible (a). PNS View showed complete opacification of the left maxillary sinus (star). Destruction of the postero-lateral wall was seen on the left side (red arrows) as opposed to the contralateral normal side (yellow arrows) (b)

fluid-filled space occupying the left nasolabial, buccal, and masseteric regions. It had destroyed the superior alveolar margin and bony confinement of left maxillary sinus anteriorly and partly eroded the zygoma. MRI [Figure 4a and b] revealed a well-defined, irregular-shaped soft-tissue mass with central necrosis, epicenter of the mass being left masticator space. The entire left masseter and buccal spaces were involved along with pterygoid muscles with erosion of the ramus of the mandible. Anteriorly, the mass was compressing the left maxillary antrum with smooth scalloping of its wall but no invasion into the sinus. Superiorly, the mass involved the infratemporal fossa causing smooth erosion of the skull base. The mass was hypointense on T1W, and areas of heterogeneous enhancement were seen on T2W images, suggestive of schwannoma of masticator space. FNAC results also suggested a highly cellular benign spindle cell tumor with nuclear palisading. Surgery was performed under general anesthesia and the mass was resected. Histopathological examination revealed intensely hematoxiphilic nucleated spindle cells arranged parallel in a palisading manner around Verocay bodies, suggestive of Antoni A tissue [Figure 5a: 40×], and edematous connective tissue stroma, dilated capillaries, and streams of loosely packed spindle cells in the interstitium, suggestive of Antoni B type tissue [Figure 5b: 10×], overall confirming the diagnosis of schwannoma.

Discussion

Schwann cells myelinate the axons peripherally, embarking at the Redlich–Obersteiner's zone, a transitional junction between the central and the peripheral axonal myelination.[2] In head and neck schwannomas, when the nerve of origin is small, it is encapsulated. If large, the tumor generally forms inside the epineural sheath, attaching the component nerve fascicules to its capsule, but sparing the axons, displacing and compressing the normal elements of the nerve to one side. This pathogenesis implies that surgical excision can preserve the nerve of origin by separating the mass from the underlying nerve fibres.[3] The tumor, in the present case, showed cystic degeneration, secondary erosion, and scalloping of the bony walls of maxilla, coronoid process and ramus of the mandible, and infratemporal fossa. However, the nerve of origin could not be traced, but was judged to arise from a branch of mandibular nerve as exhibited by the tumor's anatomic site. Locating the nerve of origin mounts to a herculean task, wherein direct relation with the nerve cannot be demonstrated in approximately 10%–40% of the cases.[2,4]

Nakamura *et al.* reported benign masseteric schwannoma as a slowly enlarging, painless cheek mass covered with normal skin, causing facial disparity, while others have reported painless masseteric nodules, physical discomfort, trismus, and limitation in jaw movement, findings similar to this case.[5,6] Mahajan *et al.* have reported progressively increasing paresthesia and numbness of the face in a patient with occasional headaches, ataxia, and weakness of muscles of mastication.[7] Ishikawa *et al.* reported a case of masseteric schwannoma with unilateral hemicrania, dull dragging pain, swelling of right cheek when the mouth was maximally opened, ringing in the ear, with numb sensation in the arm, and optical disturbances.[8]

Relatively uncommon in children, these neoplasms commonly occur between the ages of 20 and 50 years with a female predilection. Intraorally, the most common anatomical site is the tongue, followed by the palate, floor of the mouth, buccal mucosa, gingiva, and lip.[9] Grossly, schwannomas can be divided into intraosseous and peripheral tumors. Small schwannomas are relatively solid, but the bigger ones exhibit degeneration in the center due to vascular thrombosis and subsequent necrosis, thereby becoming partly cystic as seen in the present case.[10] Differential diagnosis must be established from other clinically similar benign processes such as masseteric hypertrophy, hemangiomas, neuromas, neurofibromas, granular cell myoblastoma, neuroepitheliomas, fibromas, or adenomas.[9]

Histopathologically, Antoni A type tissue shows compactly arranged spindle-shaped cells, with long oval or rod-shaped nuclei, that are frequently oriented with their long axes parallel to one another, creating a pattern of palisades, within the cytoplasmic matrix. This alignment of nuclei in rows, separated by clear acellular amorphous hyaline bands along the longitudinally cut bundles, is called Verocay bodies. Antoni B type tissue have a hypocellular pattern lacking in arrangement of bundles or palisades, portraying polymorphic tumor cells, and dispersed in

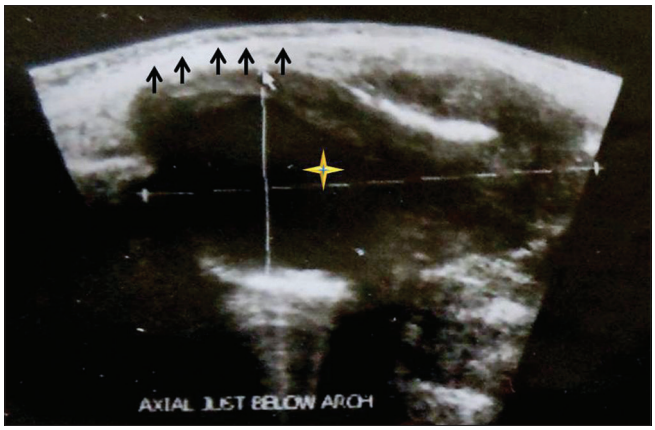

Figure 3: Ultrasonogram revealed a hyperechoic, thick-walled, soft tissue mass (black arrows) with a central hypoechoic paisley-shaped, fluid-filled area (yellow star) below the zygomatic arch, occupying the buccal and masseteric regions

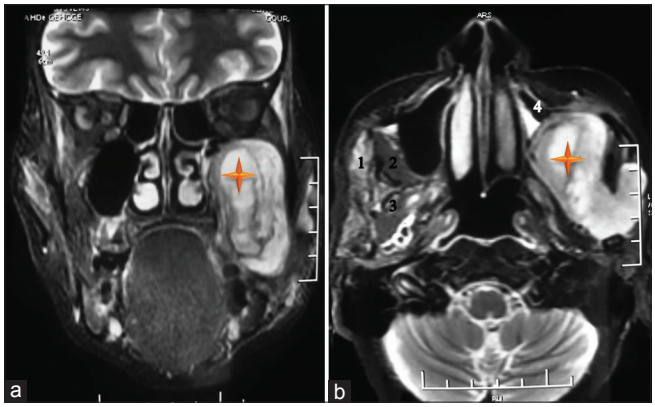

Figure 4: Coronal (a) and axial (b) sections of contrast-enhanced T2W MRI revealed hyperintense, soft-tissue mass with central necrosis, epicentre of the mass being left masticator space (yellow star). Left masseter and buccal spaces were involved alongwith pterygoid muscles with erosion of the ramus of the mandible (1,2, and 3 represent the normal masseteric and buccal spaces, and pterygoid muscles on the contralateral right side). Mass compressing the left maxillary antrum (4) seen with smooth scalloping of its wall

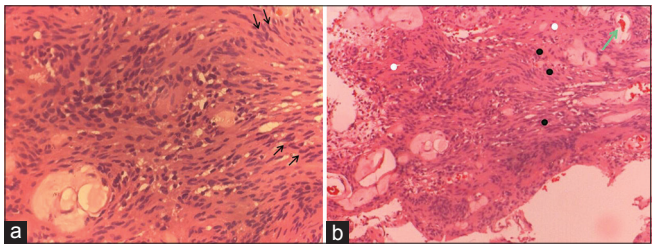

Figure 5: Intensely hematoxiphilic nucleated spindle cells arranged parallel in a palisading manner around Verocay bodies (black arrows; a: ×40) suggestive of Antoni A tissue (black dots), and oedematous connective tissue stroma, dilated capillaries (green arrow), and streams of loosely packed spindle cells in the interstitium, suggestive of Antoni B type tissue (white dots; b: ×10)

abundant eosinophilic matrix. Mucinous and microcystic changes occur more often in type B tissue.[3] Immunohistochemistry of these tumors shows diffuse, strong nuclear positivity for S-100, CD 34, and capsular epithelial membrane antigen.[9]

MRI is more sensitive and specific than computed tomography scan for schwannoma. MRI allows accurate measurement of tumor size and precise localization in identifying the tumor and its correlations with surrounding vascular structures, muscles, and nerves. T1-weighted images are hypointense to intermediate signals, whereas T2-weighted images are hyperintense. Images can be both heterogeneous and homogeneous and are usually well enhanced after gadolinium injection.[11] Nonenhancing cystic or necrotic areas are typically found in large schwannomas.[10] In treatment protocol, preference is usually given to intracapsular enucleation over complete tumor resection, with special attention to preserving the nerve of origin along with lymphovascular structures.[3,12]

Conclusion

The masticator space commonly exhibits infection and inflammatory conditions. However, the fascial compartment can be invaded by neoplasm from adjacent structures or from hematogenous matastases. In general practice, most of the patients usually visit community-based primary care physician or dental personnel when observing any growth or swelling in the oral cavity. They tend to avoid expensive and overloaded hospital facilities. It is the duty of such physicians to not only make pocket-friendly diagnostic and treatment decisions but also a judicious investigation and referral call when suspecting an intraoral tumor with a malignant potential. They should attend more continuing medical education symposia, conferences, and workshops to update knowledge and promote early screening and diagnoses of tumors and cancer. In this way, early detection is feasible thereby alerting the patient and prompting a definitive preoperative diagnosis assisted with suitable imaging and biopsy. Early intervention prevents malignant transformation of the tumor and preserves the structural and functional integrity of the nerve concerned.

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Conflicts of interest

There are no conflicts of interest.

References

1. Joshi R. Learning from eponyms: Jose Verocay and Verocay bodies, Antoni A and B areas, Nils Antoni and Schwannomas. Indian Dermatol Online J 2012;3:215-9.
2. Redman RS, Guccion JG, Spector CJ, Keegan BP. Cellular schwannoma of the mandible: A case report with ultrastructural and immunohistochemical observations. J Oral Maxillofac Surg 1996;54:339-44.
3. Parizel PM, Geniets C. Tumors of peripheral nerves. In: DeSchepper AM, Vanhoenacker F, Gielen J, Parizel PM, editors. Imaging of Soft Tissue Tumors, 3rd ed. Berlin, Heidelberg, New York: Springer; 2006. p. 325-53.
4. Pfeifle R, Baur DA, Paulino A, Helman J. Schwannoma of the tongue: Report of 2 cases. J Oral Maxillofac Surg 2001;59:802-4.

5. Nakamura A, Iguchi H, Kusuki M, Zushi T, Yamane H, Wakasa K. Intramasseteric schwannoma in a child. Auris Nasus Larynx 2006;33:347-50.
6. He Y, Fu HH, He J, Zhu HG, Zhang ZY. Schwannoma arising from intramasseteric region. J Craniofac Surg 2010;21:1998-2001.
7. Mahajan A, Sable NP, Vaish R, Moiyadi A, Arya S, D'Cruz AK. Multicompartmental trigeminal schwannomas: Dumbbell tumors revisited. J Glob Oncol 2016;2:431-5.
8. Ishikawa T, Murakami K, Yasui R, Maeda K, Tanaka K, Shimosato T. A multifocal schwannoma of the masseteric nerve causing hemicrania. Oral Surg Oral Med Oral Pathol 1991;71:329-32.
9. Nayyar AS, Khan M, Vijayalakshmi KR, Hemavathy S, Sampath S. A Benign tumor masquerading malignant behavior. Int J Oral Maxillofac Pathol 2012;3:55-9.
10. Stout AP. Tumors of peripheral nervous system: Atlas of tumor pathology, Sect 2, fasc 6. Washington, DC: The Armed Forces Institute of Pathology; 1949.
11. Liu HL, Yu SY, Li GK, Wei WI. Extracranial head and neck schwannomas: A study of the nerve of origin. Eur Arch Otorhinolaryngol 2011;268:1343-7.
12. 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;34:44-50.