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Benign neurilemmoma in the infratemporal fossa involving maxillary sinus and pterygopalatine fossa

Jin-Woo Choi, Min-Suk Heo, Chang-Hyeon An*, Jin-Koo Lee, Won-Jin Yi, Sam-Sun Lee, Soon-Chul Choi

Department of Oral and Maxillofacial Radiology, Dental Research Institute, and BK21, College of Dentistry, Seoul National University
**Department of Oral and Maxillofacial Radiology, College of Dentistry, Kyungpook National University*

ABSTRACT

Neurilemmoma is a benign tumor of the nerve sheath that arises on cranial and spinal nerve roots as well as along the course of peripheral nerves. A case of a neurilemmoma that arose in the left infratemporal fossa of a 29-year-old male was presented. Plain radiographs, enhanced computed tomography scan, and magnetic resonance imaging demonstrated a large, well-circumscribed, heterogeneously enhanced mass with extension into the pterygopalatine fossa. Displaced by the large mass, bowing-in of the posterior maxillary antral wall was noted and a provisional diagnosis of a benign soft tissue tumor was made. The mass was completely excised and a diagnosis of neurilemmoma was confirmed. (***Korean J Oral Maxillofac Radiol 2004; 34 : 215-8***)

KEY WORDS : Neurilemmoma; Head and Neck Neoplasms; Magnetic Resonance Imaging

Verocay who named them neurinomas first established neurilemmoma as a pathologic entity in 1910.[1] Stout coined the term neurilemmoma, believing that this tumor arose from the cells of the sheath of Schwann.[2] Previously; this tumor had once been designated as schwannoma, encapsulated neurofibroma, and perineural fibroblastoma.

It usually occurs as a solitary soft tissue or intrabony lesion, which is slow growing, encapsulated, and often with the associated nerve attached peripherally.[2] This is in contrast with the other benign nerve sheath tumor, the neurofibroma, which arises not only from the nerve sheath, Schwann cells but also other enveloping membranes of the trunk.

Histopathologically, neurilemmoma is a unilocular mass surrounded by a capsule of epineurium and residual nerve fibers, often with the peripheral nerve attached to the edge of the neoplasm. The substance of the tumor is composed of a mixture of two cellular patterns, namely Antoni type A and B. In contrast to the neurofibroma, which contains a mixture of various cell types, the neurilemmoma consists almost exclusively of Schwann cells.[2-5]

Usually, the mass is asymptomatic, although tenderness or pain may occur in some instances. The lesion is most common in young and middle-aged adults.[3,6] It has been reported, that no gender differences of distribution exists.[7] Neurilemmoma has been reported in almost every part of the body and is relatively uncommon, but 25 to 48 percent of all cases occur in the head and neck region.[3] Among those, there were few reports of neurilemmoma arising primarily from the infratemporal space.[6,8]

We present this case because of its unusual location in the infratemporal fossa and severe anterior bowing-in of the maxillary sinus wall.

Case report

A 29-year-old man with slight left facial swelling was referred to our department from other dental hospital after computed tomography (CT) taking in January 2004. The left upper third molar was extracted at a local clinic 2-years ago and he has been feeling slight mobility of adjacent teeth since then. There had been no history of pain or other associated symptoms except the tooth mobility.

In plain radiographs, the posterior wall of left maxillary sinus was displaced anteriorly close to anterior wall and orbital floor by the mass (Fig. 1). Root resorption of the left maxillary second premolar, first molar and second molar was shown.

CT scans demonstrated a discrete, well-circumscribed, heterogeneously enhanced mass. The main mass was located in the infratemporal fossa area. However, the mass was so enormous that the posterior wall of the left maxillary sinus was collapsed anteriorly and the pterygoid plate was displaced posteriorly and the floor of orbit was thinned by the mass (Fig.

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Correspondence to : Prof. Sam-Sun Lee
Department of Oral and Maxillofacial Radiology College of Dentistry, Seoul National University 28, Yeongeon-dong, Jongno-gu, Seoul, 110-749, Korea
Tel) 82-2-760-3498, Fax) 82-2-744-3919, E-mail) raylee@snu.ac.kr

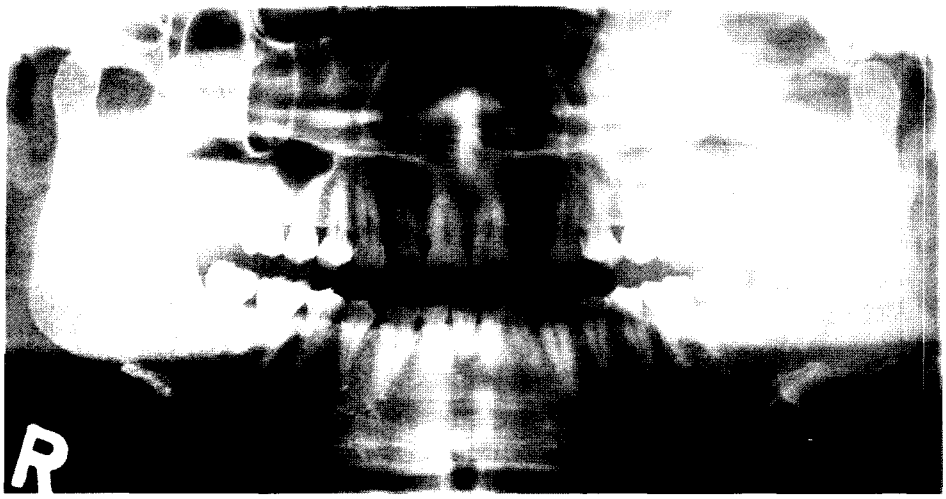

Fig. 1. Panorama shows bowing-in of the left posterior antral wall and root resorption.

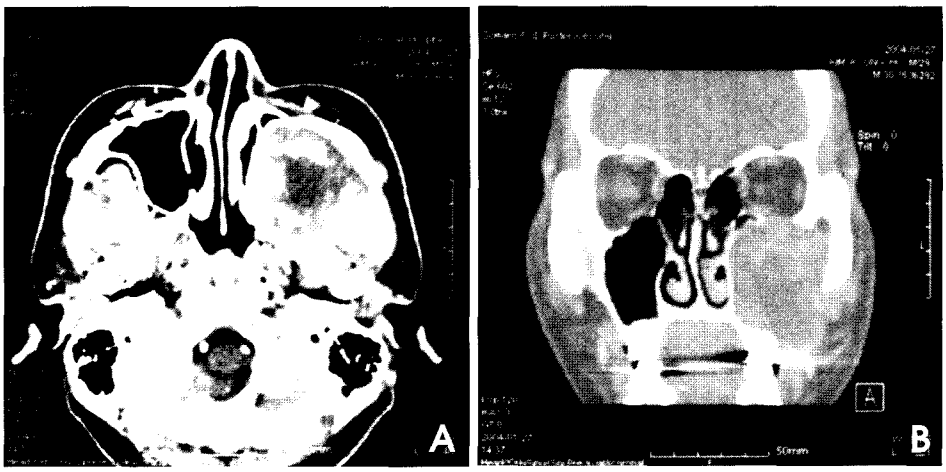

Fig. 2 A: Axial enhanced CT shows bowing-in of the left maxillary sinus posterior wall and displacement of the pterygoid plate. B: Coronal CT shows displacement of the nasal wall and thinning of the orbital floor.

2A, B). The sphenoid sinus, nasal cavity, and pterygopalatine fossa were also affected. A benign tumor was suspected when bowing of the posterior maxillary antral bony wall was observed with continuity on CT scan and plain radiographs.

In the magnetic resonance (MR) images, T1 weighted image demonstrated a well circumscribed, low signal intensity extra-antral mass and compressing the maxillary sinus posterior wall anteriorly (Fig. 3A). The mass was separated into central and peripheral portion on every MR sequences, by low signal intensity rim, which may indicate fibrotic tissue of the neurilemmoma. In the contrast enhanced T1 weighted image, central portion was not enhanced while peripheral portion showed heterogeneous enhancement (Fig. 3B). In the fat-suppressed T2 weighted image, central portion showed high signal intensity but not as high as water signal intensity, whereas peripheral portion demonstrated intermediate signal intensity (Fig. 3C). The extension along the pterygopalatine fissure to the pterygopalatine fossa was observed (Fig. 3D and E).

The tumor was enucleated as a single mass and biopsy was done (Fig. 4). The immunohistochemical stain disclosed a positive reaction to S-100 protein. Neuron specific enolase (NSE) and Glial fibrillary acidic protein (GFAP) were also positive and a diagnosis of neurilemmoma was confirmed. The only postoperative problem of the patient was diminished sensation of left maxillary area. However, the enhanced region of the operation site was noted on follow-up MRI taken at three months after surgery recall check up, and the patient is on the process of follow-up.

Discussion

Unusually this tumor occurred in the infratemporal fossa, showed evidence of originating from the second division of the fifth cranial nerve near the foramen rotundum. Clinically neurilemmoma presents with the symptoms related to the nerve from which it arises. Pain associated with a mass is the

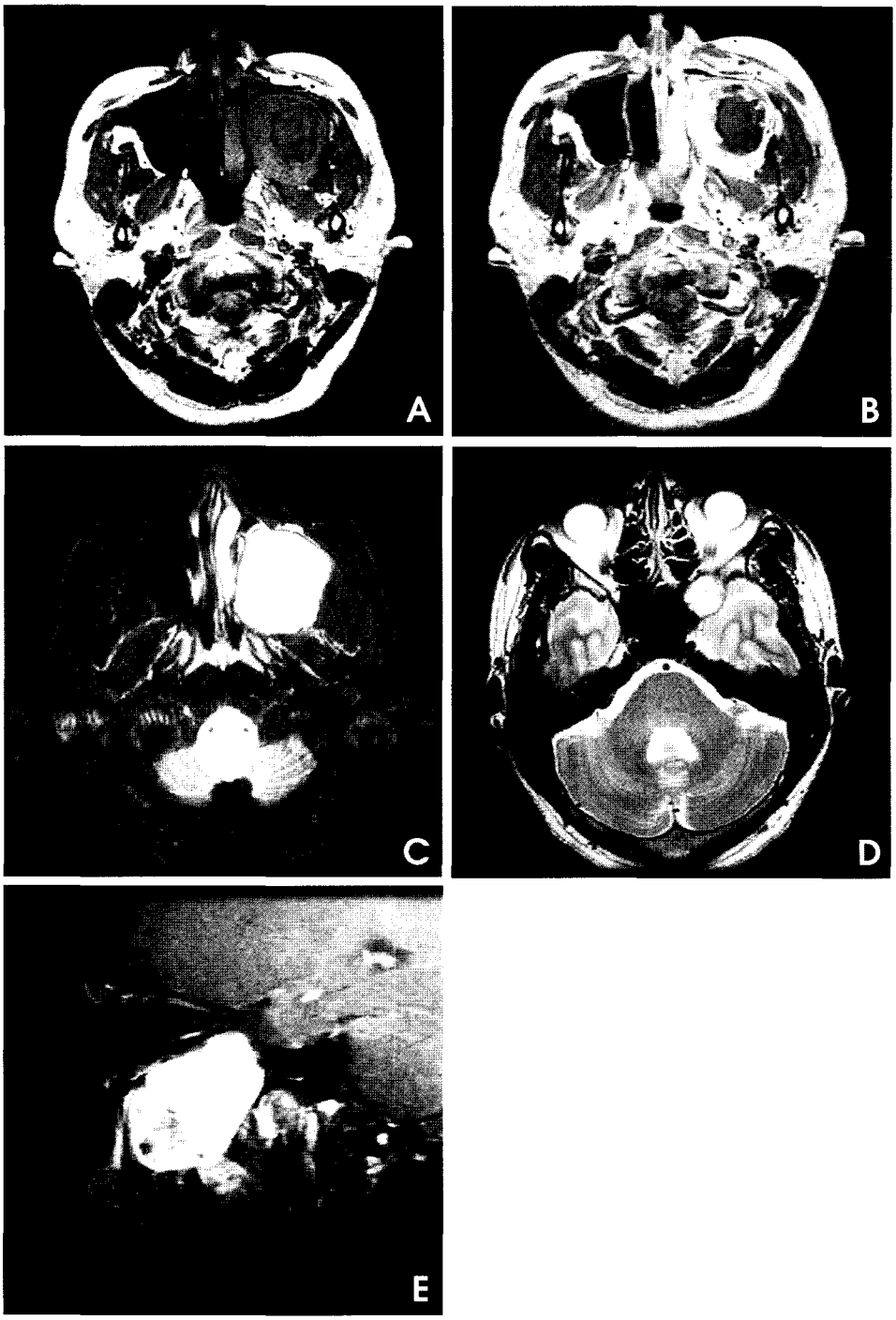

Fig. 3. A: T1 weighted image shows low signal intensity of central and peripheral part. B: Contrast enhanced T1 weighted image shows non-enhancing central portion and heterogeneous enhancing peripheral part. C: Fat-suppressed T2 weighted image shows slight high signal intensity of central portion and intermediate intensity of peripheral portion. D: and E: T2 weighted image shows tumor expansion along the pterygopalatine fissure and pterygopalatine fossa involvement.

most common presenting symptom of neurilemmoma and other symptoms could be hearing loss, hoarseness and dysphagia, depending on the site of involvement.[9] However, lack of symptom is common when the lesion is originated with the trigeminal nerve and located in the infratemporal area. In our case, extension to the pterygopalatine fossa indicates the possibility of the tumor originating from the second division of trigeminal nerve (Fig. 3D and E).

Neurilemmoma is a benign tumor of the nerve sheath that arises on cranial and spinal nerve roots as well as along the course of peripheral nerves. Intracranial neurilemmoma is relatively common. It accounts for about 8% of all primary tumors in this region.[8] Among these, vestibulo-cochlear neurilemmoma is the most common and has been the subject of extensive investigation.[10] Neurilemmoma of the trigeminal nerve accounts for approximately 0.2% of intracranial tumors

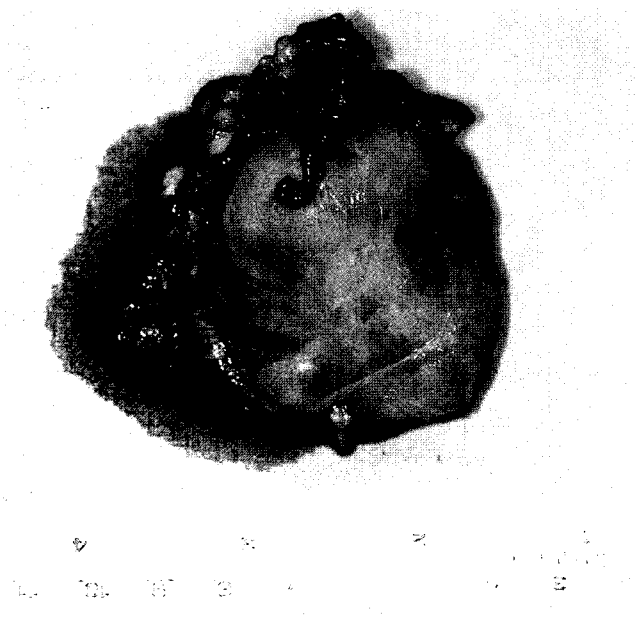

Fig. 4. Photograph shows the excised specimen.

and 2% of all intracranial neurilemmoma.[11] Of all extracranial sites of neurilemmoma, the head and neck region is the most common site.[2,3,12,13] The incidence has been reported to be 37 to 45 percent of all solitary neurilemmoma.[3,12] In the head and neck area, neurilemmoma most frequently occurred in the parapharyngeal area in association with lower cranial nerves or the sympathetic chain.[14] The infratemporal fossa is one of the least common anatomical sites for neurilemmoma.[6,8]

Most neurilemmomas do not reach a large size. In fifty cases reported by Stout,[2] only five tumors were measured to be 6 cm or more in diameter. The average size of tumor mass was less than 5 cm,[3] and only rarely did they exceed 10 cm.[6] However the infratemporal fossa is a specific anatomic region situated deep to the ramus of the mandible. Tumors growing in this clinically silent area may reach considerable size before producing symptoms or becoming evident on clinical examination.[15] The tumor in our article was measured 49 × 47 × 58 mm on MR scan and caused severe anterior antral bowing.

Holman and Miller[16] first denoted bowing of the posterior maxillary antral wall as a characteristic sign of juvenile nasopharyngeal fibroma in 1965. It can be clearly seen on plain radiograph and CT scan. However, Som et al.[17] showed that the antral bowing sign is not characteristic for juvenile nasopharyngeal fibroma alone, but can also reflect the presence of a slow-growing mass in the retromaxillary space. Our article and a few cases of neurilemmoma originating from infratemporal space have shown the antral bowing sign.[18,19]

We present here a benign neurilemmoma in the infratemporal fossa which is enlarged very much without any specific symptom, and we look forward that this report will add more radiographic information for diagnosis this pathosis.

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