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Ancient schwannoma in the parotid gland: A case report and review of the literature

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ABSTRACT

Schwannomas are uncommon neoplasms of neurologic origin that are rare in the salivary glands. A schwannoma that persists for a long time is referred to as an ancient schwannoma if it is accompanied by degenerative changes on histology. The case described herein involved a 37-year-old man with an ancient schwannoma that had persisted for 20 years in his right parotid gland. Clinically, the lesion presented with swelling and pain. Computed tomography revealed a well-defined, multilocular enhanced lesion. T2-weighted magnetic resonance images displayed multilocular hyperintensity, while T1-weighted images showed a high signal at the lobulated margin and a homogeneous low signal internally. The preoperative diagnosis, based on the lesion's location and imaging diagnosis, was Warthin's tumor. However, a biopsy conducted after surgical excision identified the lesion as a schwannoma with cystic degeneration. This report also presents a summary of the characteristics of rare cases of schwannoma in the major salivary gland based on this case and a literature review. ***(Imaging Sci Dent 2023; 53: 239-45)***

KEY WORDS: Neurilemmoma; Parotid Gland; Magnetic Resonance Imaging; Tomography, X-ray Computed

Introduction

Schwannomas, also known as neurilemmomas, are neurogenic benign neoplasms derived from Schwann cells, which encapsulate nerves in the peripheral nervous system. This condition was first described by Verocay in 1910.[1] These lesions are encapsulated, slow-growing tumors that are associated with adjacent nerve bundles and rarely undergo malignant transformation.[2] They can be clinically detected when they exert pressure on relevant nerves or structures, and typically present minimal symptoms until they invade adjacent tissue.[2] Microscopically, schwannomas can be roughly divided into those in Antoni A areas, which have a high density of spindle cells, and those in Antoni B areas, which are mainly composed of myxoids.[3] Histologically, schwannomas can be divided into 5 subgroups: common, plexiform, cellular, epithelioid, and ancient.[4] The term "ancient schwannoma" was first introduced by Ackerman and Taylor.[5] This variant is characterized by long-standing degenerative changes, including decreased cellularity, partial hyaline accumulation, tissue fat alteration, calcification, and hemorrhage.[5] If the lesion persists for an extended period, these degenerative changes may result in the predominant presence of cystic myxoids, accompanied by spindle cell deformation.

Although 45% of schwannomas occur in the head and neck area, they very rarely develop in the salivary gland.[7] Benign tumors of the salivary gland account for approximately 3% of all tumors, with 80% of these found in the parotid glands.[2,8] The occurrence of a schwannoma within these parotid tumors which involved an ancient schwannoma that had undergone degeneration with cystic changes over an extended period is extremely rare.[9]

This report presents a rare case of ancient schwannoma in the parotid gland, along with a literature review of schwannomas in the major salivary gland.

Case Report

A 37-year-old man presented to the Department of Oral and Maxillofacial Surgery, Kyungpook National University Dental Hospital, with a mass in the right preauricular region that had been growing progressively for 20 years. Although

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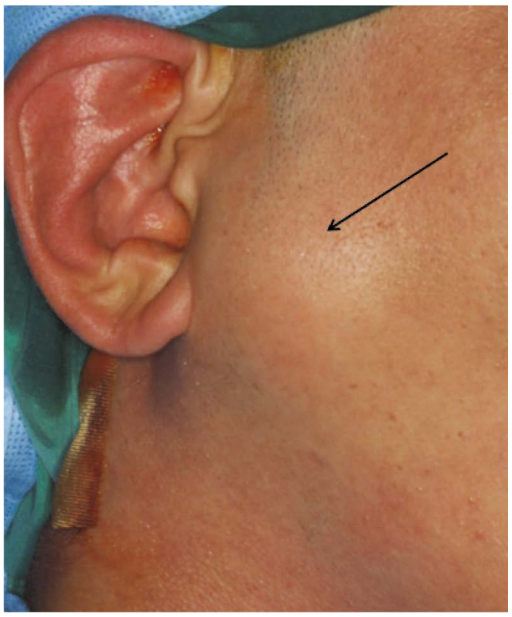

Fig. 1. Clinical photograph shows a round mass occupying the right preauricular region (arrow).

there were no specific symptoms, the size of the nodule had begun to increase gradually over the past 2 years, accompanied by pressure pain. A physical examination revealed no tenderness, fluctuation, or pain upon palpation of the right parotid region, but a mass measuring 3 cm in diameter was detected (Fig. 1).

Panoramic radiography (Orthopantomograph OP 200D, Instrumentarium Dental, Tuusula, Finland) showed no bony changes associated with the right condylar head or its surrounding structures (Fig. 2). Contrast-enhanced computed tomography (CT) (Optima CT660, GE Healthcare Co, Milwaukee, WI, USA) revealed a well-defined multilocular mass with peripheral enhancement and low internal density in the superior-anterior portion of the right parotid gland (Fig. 3). T1-weighted magnetic resonance (MR) imaging (Signa HDxt 3.0 T, GE Healthcare Co, Milwaukee, WI, USA) showed a well-defined ovoid hypointense and homogeneous lesion with an internal signal isointense to the muscle. In contrast, corresponding T2-weighted MR images

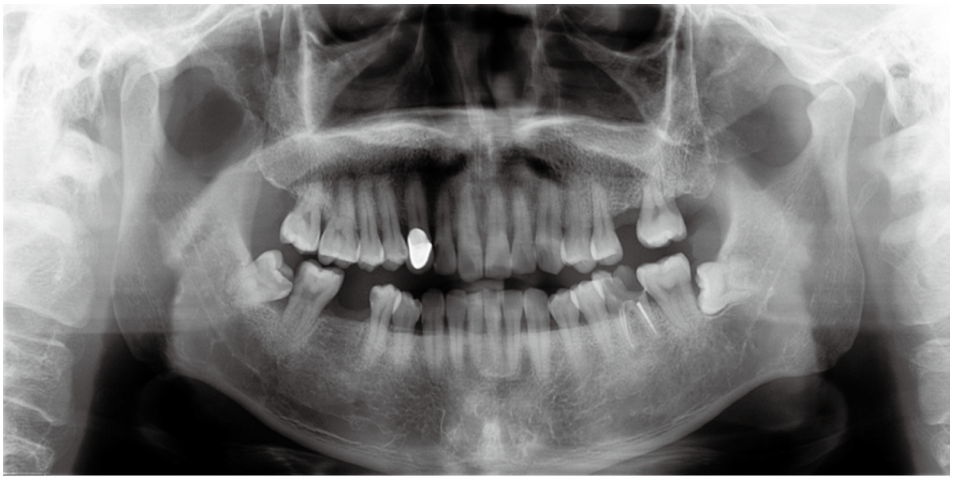

Fig. 2. Panoramic radiograph shows no pathological changes associated with the lesion in the right parotid gland area.

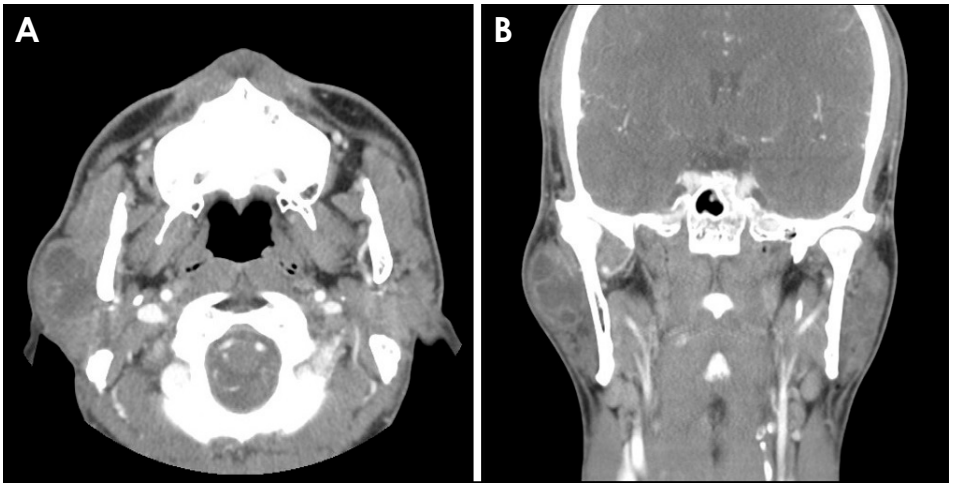

Fig. 3. Axial (A) and coronal (B) contrast-enhanced computed tomography scans show an encapsulated and homogeneously enhanced multilocular soft tissue mass on the right parotid gland.

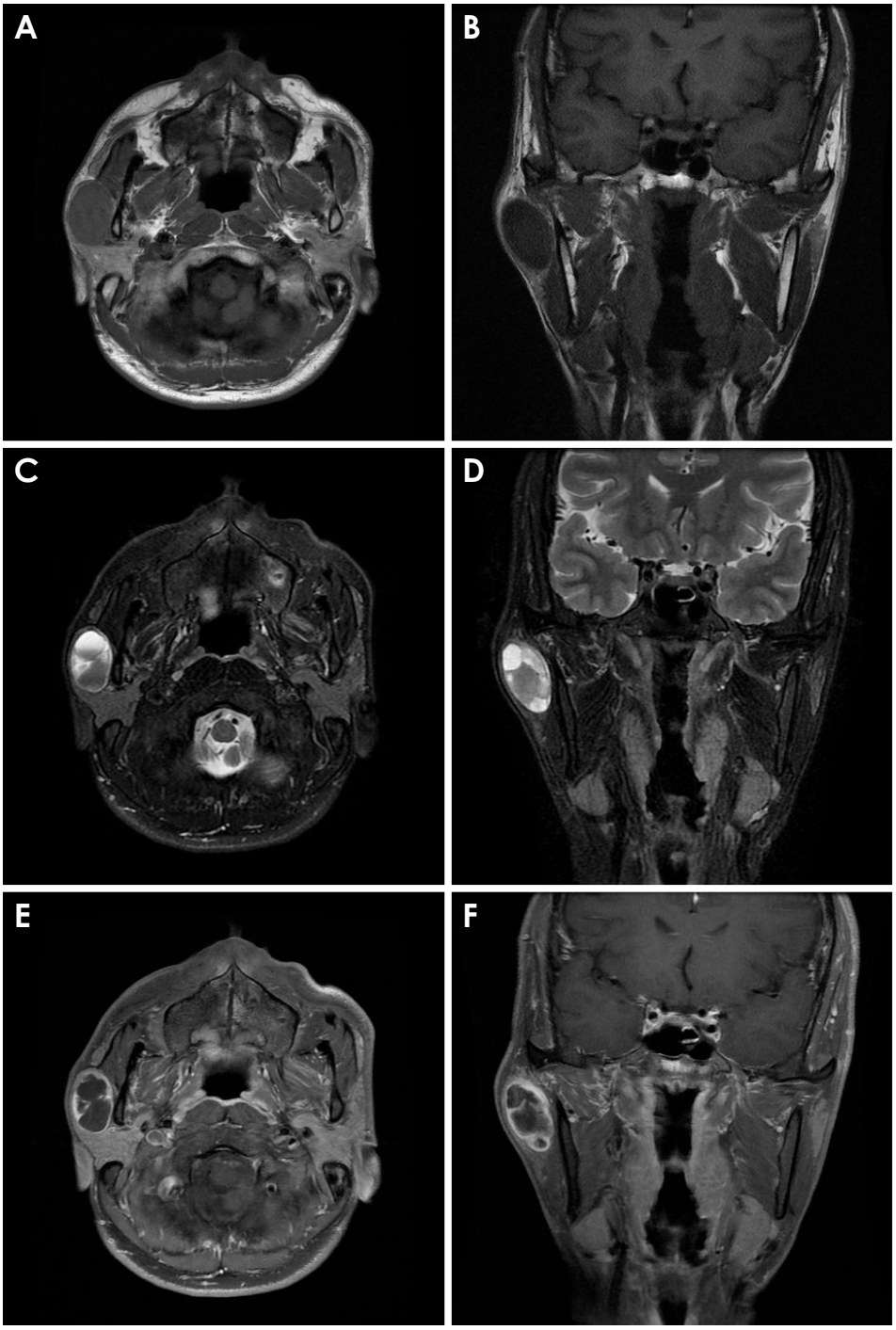

Fig. 4. Axial (A) and coronal (B) T1-weighted magnetic resonance (MR) images show a well-defined homogeneous hypointense mass of the right parotid gland area. Axial (C) and coronal (D) T2-weighted MR images show the ovoid mass with multilocular hyperintensity and a partially hypointense signal. Axial (E) and coronal (F) fat-suppressed enhanced T1-weighted MR images show the lesion with a high signal along the lobulated margin and an internal homogeneous low signal.

displayed a well-circumscribed tumor with a signal hyperintense to the muscle and internal multilobular high signal due to cystic fluid content. On a fat-suppressed enhanced T1-weighted MR image, the margin and lobulated section of the tumor exhibited a higher signal than the remaining gland, and the internal structure displayed a homogeneous low signal (Fig. 4).

A fine needle aspiration biopsy (FNAB) was performed prior to surgery, but it did not yield any information useful for preoperative diagnosis. Tentatively, these radiologic and clinical features suggested a Warthin's tumor in the parotid gland. The lesion was surgically excised under general anesthesia. Tissue dissection was carried out through a modified Blair incision, and the tumor was examined. The facial nerve branches were preserved and dissected from the tumor, with particular attention paid to the right buccal branch of

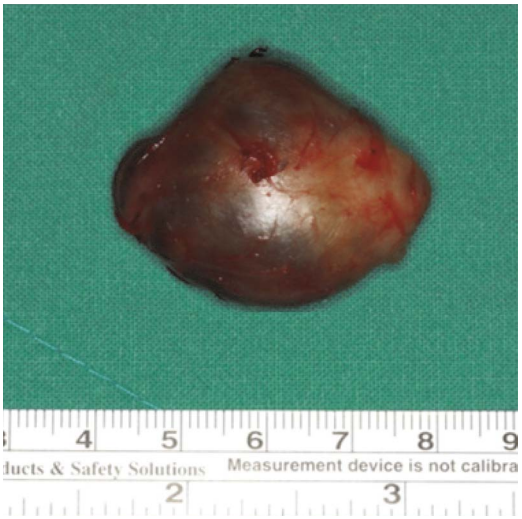

Fig. 5. Surgical specimen measuring 3.9 cm × 2.9 cm in diameter shows the encapsulated smooth mass.

the facial nerve. A nerve stimulator was also utilized during the operation. The excised lesion measured 3.9 cm × 2.9 cm (Fig. 5).

Histopathological analysis revealed several large and small cystic changes suggestive of degenerative changes in the tumor. Spindle-shaped Schwann cells with a palisaded arrangement (Antoni A) and spindle cells with loose myxoid stroma (Antoni B) were also observed. Immunohistochemical findings showed that the Schwann cells positively responded to S-100 protein (Fig. 6). Consequently, the lesion was diagnosed as a schwannoma with cystic change through a postoperative biopsy.

At the postoperative follow-up, the patient experienced discomfort while winking his right eye for a week. However, he subsequently had an uneventful recovery without any facial nerve damage following the surgery. A follow-up

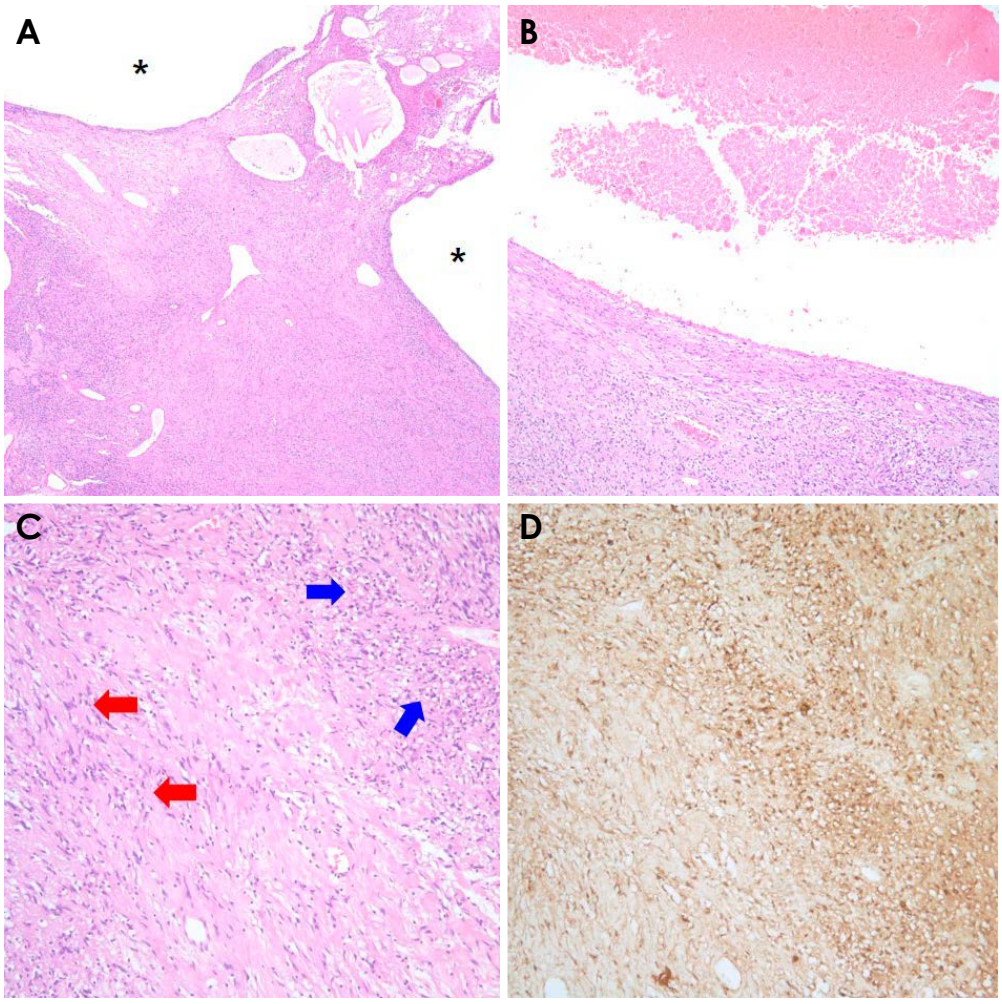

Fig. 6. A. A significant cystic change (black asterisk) and multiple small cysts inside the mass are observed (hematoxylin and eosin [H&E] stain, ×40). B. Hemorrhages are observed in the lumen (H&E stain, ×100). C. Spindle-shaped Schwann cells form a palisaded arrangement around the eosinophilic area (Antoni A, red arrow). Spindle cells are arranged with a loose and myxomatous stroma. (Antoni B, blue arrow) (H&E stain, ×200). D. Spindle-shaped cells are positive for S-100 protein (immunohistochemical stain, ×200).

examination was conducted at 2 years and 10 months, and no recurrence or discomfort was reported.

Discussion

Benign tumors of the salivary gland account for 3%-10% of head and neck tumors, with 80% of these occurring in the parotid glands.[2,8,10] The majority of benign lesions in the parotid gland are pleomorphic adenomas, followed by Warthin's tumors.[10] In the present case, the parenchymal tissue within the tumor exhibited a lobulated cystic change with clear boundaries on CT and MR imaging. Consequently, a tentative diagnosis of Warthin's tumor was made based on these imaging findings.[11] Liu et al.[12] suggested that it is not possible to distinguish between pleomorphic adenoma and Warthin's tumor due to the absence of differences in tumor margins or density on CT images.[12] An attempt was made to use FNAB for cytological diagnoses, but no useful information for preoperative diagnosis was obtained. The accurate diagnosis of a schwannoma is almost exclusively confirmed through an intraoperative tissue biopsy.[6,9,11] Since the aspiration site is operator-dependent and variable, it is challenging to make an accurate diagnosis by extracting only the spindle cells typically observed in schwannomas.[9] Therefore, it is often the case that FNAB is not very helpful in the preoperative diagnosis of schwannomas.[9]

Table 1 presents 7 cases of schwannomas in the major salivary glands, including the present case. In the literature review, schwannomas in the salivary glands were detected in patients aged between 11 and 46 years (mean age, 34 years). Of these patients, 4 were male, and 3 were female. The parotid gland was the most common site (4 cases), followed by the submandibular gland (3 cases). Four cases were found on the right side and three cases were located on the left side. There were no instances of simultaneous occurrence on both sides (i.e., all cases were unilateral). Upon taking the patients' histories, the duration of the lesion was found to vary from 6 to 240 months, with an average of 35.1 months. A comparison of the radiographic features of the cases revealed no imaging characteristics unique to schwannoma. Three cases were diagnosed as benign salivary gland tumors. The tumor size was typically measured between 30 mm and 51 mm. In the current case, FNAB was attempted for cytological diagnosis, but it did not yield any information useful for preoperative diagnosis. According to this review, FNAB was performed on 5 patients, with only 1 being diagnosed with schwannoma. All 7 cases of salivary gland schwannomas, including the current case, exhibited cystic change and were ultimately diagnosed as ancient schwannoma.

Schwannomas can originate from any cranial or spinal nerve, with the exception of the optic (cranial nerve [CN]

Table 1. Literature review of major salivary gland ancient schwannoma

Author (year)	Age/Sex	History	Chief complaints	Site	Nerve	Radiographic feature	Size (mm)	FNAB (diagnosis)	Initial diagnosis	Cystic change
Bondy et al. (1996)[18]	23/F	6 months	Swelling	Left SMG	Lingual Hypoglossal Mandibular	NS	30×30×40	Yes (ND)	NS	Yes
Jayaraj et al. (1997)[2]	42/M	10 years	Slow-growing mass	Left PG	Facial	Well-defined homogeneous rounded lesion (CT)	30	Yes (ND)	PA	Yes
Wakoh et al. (2005)[1]	46/F	NS	Swelling	Right SMG	Lingual	Well-defined homogeneous rounded lesion with irregular margin (CT)	30	Yes (ND)	Benign tumor	Yes
Lanteri et al. (2005)[19]	43/M	6 months	Slow-growing mass	Right PG	Facial	Irregular borders of nodule (Ultrasonography)	29×20×17	NS	NS	Yes
Ho et al. (2017)[20]	36/M	2 years	Swelling	Left SMG	Facial Hypoglossal	Well-defined cystic lesion (CT)	40×30	NS	NS	Yes
Goswami et al. (2020)[8]	11/F	5 years	Slow-growing mass	Right PG	Facial	Well-defined heterogeneous lesion (CT)	51×43	Yes (Sch.)	Sch	Yes
Current case (2017)	37/M	20 years	Swelling and pain	Right PG	Facial	Well-defined multilocular enhanced lesion (CT)	39×29	Yes (ND)	WT	Yes

NS: not stated, ND: nondiagnostic, M: male, F: female, PG: parotid gland, SMG: submandibular gland, CT: computed tomography, Sch: schwannoma, PA: pleomorphic adenoma, WT: Warthin's tumor, FNAB: fine needle aspiration biopsy

II) and olfactory (CN I) nerves, which lack Schwann cells.[2] Approximately 20%-30% of all schwannomas are found in the head and neck area, with the auditory nerve (CN VIII) being the most common site. This is followed by the vagus nerve (CN X) and the hypoglossal nerve (CN XII).[2,13] In the present case, the schwannoma originated from the facial nerve (CN VII) and penetrated the parotid gland, which is rare.[14] Specifically, only about 9% of all facial nerve schwannomas are known to occur within the parotid gland.[16] Furthermore, intra-parotid facial nerve schwannomas are exceedingly rare, representing only 0.2%-1.5% of all facial nerve tumors found within the parotid gland.[15] In a retrospective study of 3,722 schwannomas over a 38-year period, Caughey et al.[17] found that only 29 (0.7%) were associated with the facial nerve, and of these, just 8 were linked to the parotid gland.

Schwannoma is treated through tumor excision in most cases.[16] However, Marchioni et al.[15] classified schwannoma into 4 types based on the extent of damage to the facial nerve and its associated nerves during surgery for facial nerve-related schwannoma. In type A, only the tumor tissue is excised, with no damage to the nerves. In contrast, type B involves the removal of the tumor tissue along with a peripheral branch or distal division of the facial nerve. In type C, the main trunk of the facial nerve is sacrificed, while in type D, damage occurs to one or more branches, including the main trunk of the facial nerve.[15] In this particular case, only the tumor tissue was successfully removed during surgery without sacrificing the facial nerve and its branches, thus classifying it as type A according to the classification of Marchioni et al.[15] After surgery, uneven recovery was observed, but there was no permanent damage to the nerve function.[15]

It is clinically necessary to differentiate schwannoma from lipoma, fibroma, and neurofibroma, as is distinguishing it from salivary gland tumors when it is associated with the salivary gland.[10] However, it is often challenging to differentiate schwannoma from pleomorphic adenoma and Warthin's tumor, which commonly occur in the salivary gland, even when using precise diagnostic imaging techniques such as CT and MR imaging.[12,14] This difficulty arises because the nerves associated with schwannoma are not accurately depicted on CT or MR imaging, and the internal and external image characteristics of the lesion's parenchyma are strikingly similar.[14] Preoperative diagnosis with FNAB is possible, but its accuracy can be low, depending on the location of the aspirated lesion and cell composition.[9] In most instances, accurate diagnosis of schwannoma in the parotid gland is achieved through intraoperative tissue biopsy. Therefore, when performing surgery to remove a salivary gland tumor, the possibility of schwannoma should always be considered, and efforts should be made to minimize nerve damage related to the lesion. In conclusion, an ancient schwannoma may be considered if a salivary gland tumor has a long history and is accompanied by degenerative changes within the tumor.

Conflicts of Interest: None

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