



Giant Schwannoma of the tongue accompanied by difficulty in closing the mouth and dysarthria: A case report

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ABSTRACT

Schwannomas are benign nerve sheath tumors and the most common intraoral sites are the tongue. They are usually painless and slow-growing. Here we report a case of giant schwannoma of the tongue which caused difficulty in closing the mouth and dysarthria. The patient was a 58-year-old woman. She had left a mass of the tongue untreated for approximately 30 years and came to our department with chief complaint of difficulty in closing the mouth. A 95 mm painless mass was identified at the anterior tongue. Difficulty in closing the mouth and dysarthria were caused by the giant tumor of the tongue protruding out of the oral cavity. Tissue biopsy confirmed a diagnosis of schwannoma. The tumor was completely resected under general anesthesia with nasotracheal intubation using the awake fiberoptic technique. After surgery, ability to close the mouth was regained and dysarthria was improved. Recurrence has not been observed for 9 years after surgery.

1. Introduction

Schwannomas are benign tumors arising from Schwann cells of the neural sheath, which commonly occur in the nerve in the head and neck region except for the optic and olfactory nerves [1–4]. These intraoral being tumors are most frequently found in the tongue and usually asymptomatic, painless, and slow-growing [5]. Schwannomas of the tongue cause various symptoms including sore throat, snoring, dysphagia, dysarthria, sleep apnea, dysphonia, and voice change, depending on the size and location of the tumor [2]. Here we report a case of 95 mm giant schwannoma of the anterior tongue accompanied by difficulty in closing the mouth and dysarthria.

2. Case report

A 58-year-old woman had an untreated tongue mass that was pointed out by a physician approximately 30 years ago. She visited Nara Medical University Hospital in 2008 with chief complaint of difficulty in closing the mouth due to a slowly growing mass.

On the intraoral examination, an approximately 95 mm, pedunculated, tender on palpation, and relatively mobile mass with smooth surface was identified in the anterior region of the tongue. The posterior region of the tongue was normal and the boundary around the mass was

well-defined. The anterior teeth were inclined toward the labial side, and difficulty in closing the mouth and dysarthria were caused by this giant tumor of the tongue protruding out of the oral cavity (Fig. 1). Sore throat, breathing difficulty, and swallowing disorder were not present. Oral intake was possible and dysgeusia was not observed. No lymph nodes of the neck region were palpable. The patient's past medical history was unremarkable, and her family history was also unremarkable.

Radiographic examination showed inclination, movement, and loss of the upper and lower anterior teeth, which were probably caused by the increased size of the mass. Also, alveolar bone resorption due to marginal periodontitis and hyperplasia of the left styloid process were observed (Fig. 2A, B). On the MRI imaging, the T1 weighted image showed a well-defined region with low signal intensity (Fig. 3A) and the T2 weighted image showed a 93 mm × 63 mm segmented heterogeneous region with high signal intensity (Fig. 3B). A section of the tissue was collected under local anesthesia for the histopathological examination, and histopathological diagnosis of schwannoma was given.

After careful evaluation of the clinical case, in accordance with the requirements of the patient, tumor resection was performed 6 months after the first visit. General anesthesia was induced via nasotracheal intubation using the awake fiberoptic technique, and the transoral

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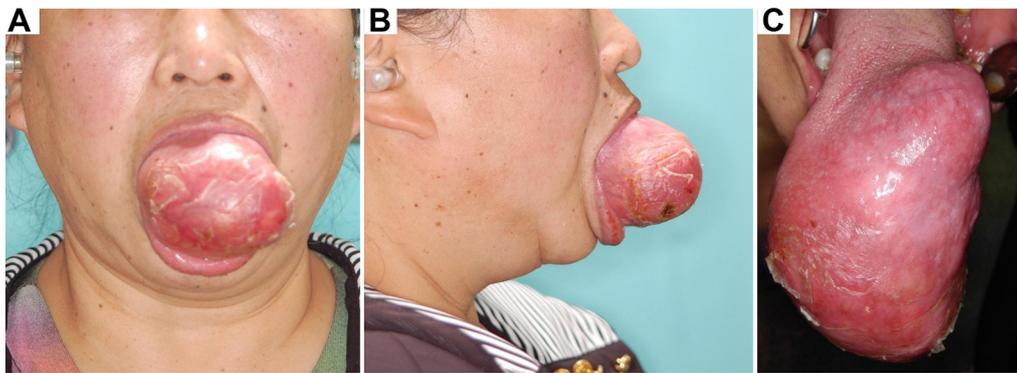


Fig. 1. Photographs of the facial appearance and tongue at the first visit. The tumor lesion protruding out of the oral cavity (A: frontal view, B: lateral view, C: tongue).



Fig. 2. Radiographs at the first visit. The inclination and loss of the anterior teeth (A: panoramic radiograph). The tongue tumor protruding out of the oral cavity and the labial inclination of the upper and lower anterior teeth (B: lateral roentgenographic cephalogram).

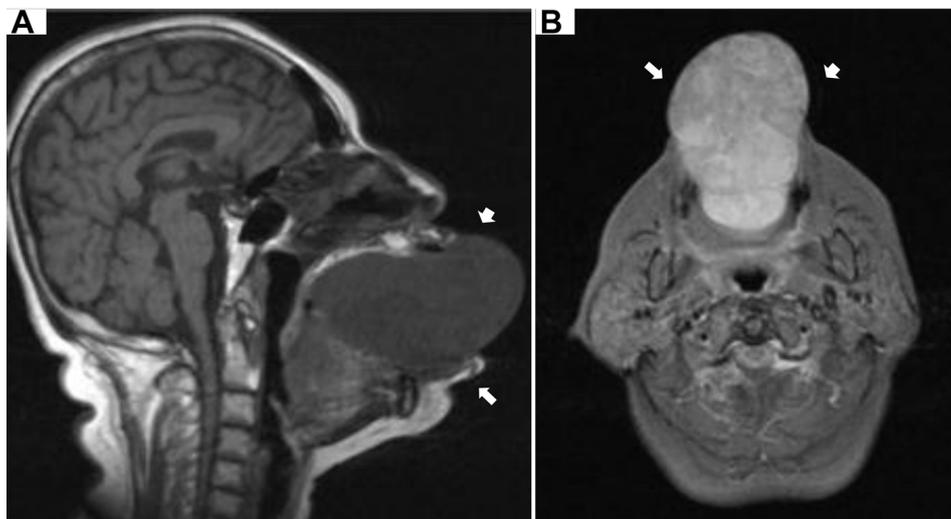


Fig. 3. MRI. A: MRI (T1 weighted sagittal image); The T1 weighted image showing a low signal lesion with clear boundary between the tumor and the surrounding tissue (white arrow). B: MRI (T2 weighted horizontal image); The T2 weighted image showing a 93 mm × 63 mm high signal heterogeneous tumor lesion with clear boundary (white arrow).

approach was selected for the surgery (Fig.4A–D). Since the tumor was clearly visualized by retracting the tongue forward, the transoral approach was selected for the surgery (Fig.4A). The tumor was resected en bloc along the capsule without adhesion to the surrounding tissue (Fig.4B). The accompanying nerve was not identified. The proglottis was reconstructed and sutured to protect tongue movement (Fig.4C, D). The resected specimen was a 95 mm × 80 mm heterogeneous solid mass with pale-yellow surface (Fig.5A, B)

The histopathological findings confirmed a relatively well-defined tumor characterized by proliferation of spindle-shaped cells and no epithelial component within the tumor (Fig.6A). The spindle-shaped cells partially showed a palisading pattern, which is known as Antoni

type A (Fig.6 B) and partially showed Antoni type B containing a myxoid stroma (Fig.6C). Furthermore, the tumor was immunohistochemically positive for S-100 protein staining (Fig.6D). The histological findings of the tumor revealed schwannoma with Antoni A and B areas.

Difficulty in closing the mouth was first improved and dysarthria was also improved soon after the surgical tumor resection. Without orthodontic treatment, the labial inclination of the teeth was improved, and the occlusal contact of the molar teeth was attained within 1 year after surgery. Radiographic examination also confirmed the improvement of the inclined teeth (Fig.7A, B). Although the anterior open bite slightly remains, the oral function was sufficiently improved with the

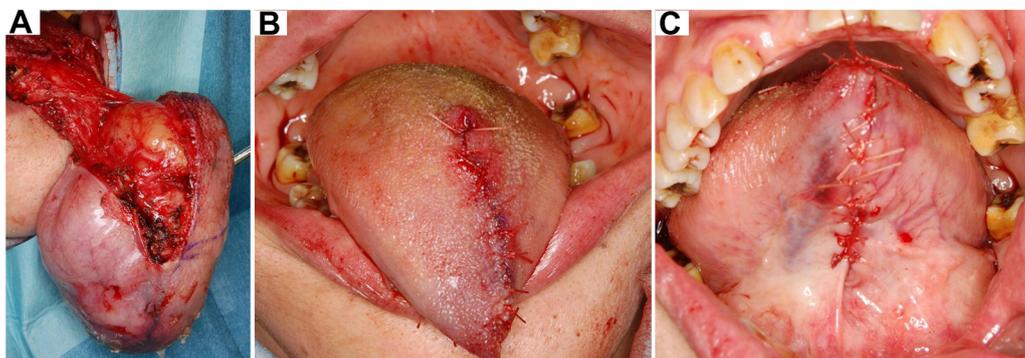


Fig. 4. Intraoperative photographs. The encapsulated and well-defined tumor of the tongue without adhesion to the surrounding tissue (A). Postoperative adequate shape and extension of the lingual frenulum attained by tongue reconstruction (B, C).

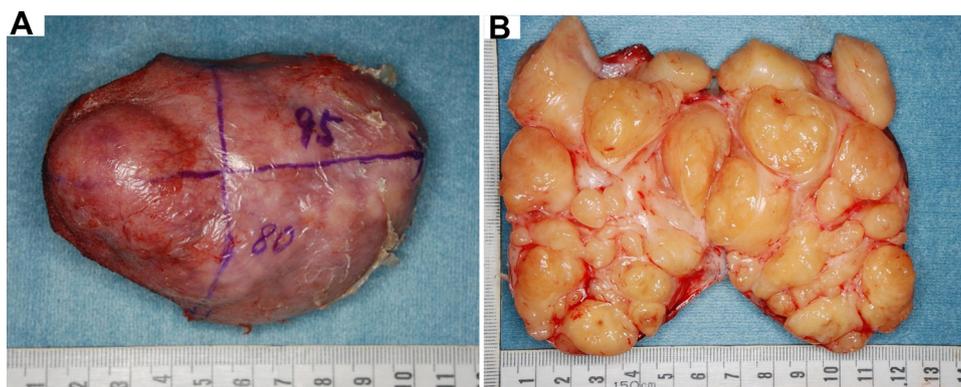


Fig. 5. Resected specimen. Whole image: The 95 mm × 80 mm tumor with smooth surface (A). Cross-sectional image: The heterogeneous solid tumor with pale-yellow surface (B).

prosthetic treatment using the denture. The sense of taste has not changed, and the sensibility and movement of the tongue have been well functioned. Recurrence has not been observed for approximately 9 years after surgery (Fig. 8A, B).

3. Discussion

Schwannoma is a benign tumor arising from Schwann cells, which was first reported as Neurinoma by Verocay in 1910. Later, Schwannoma

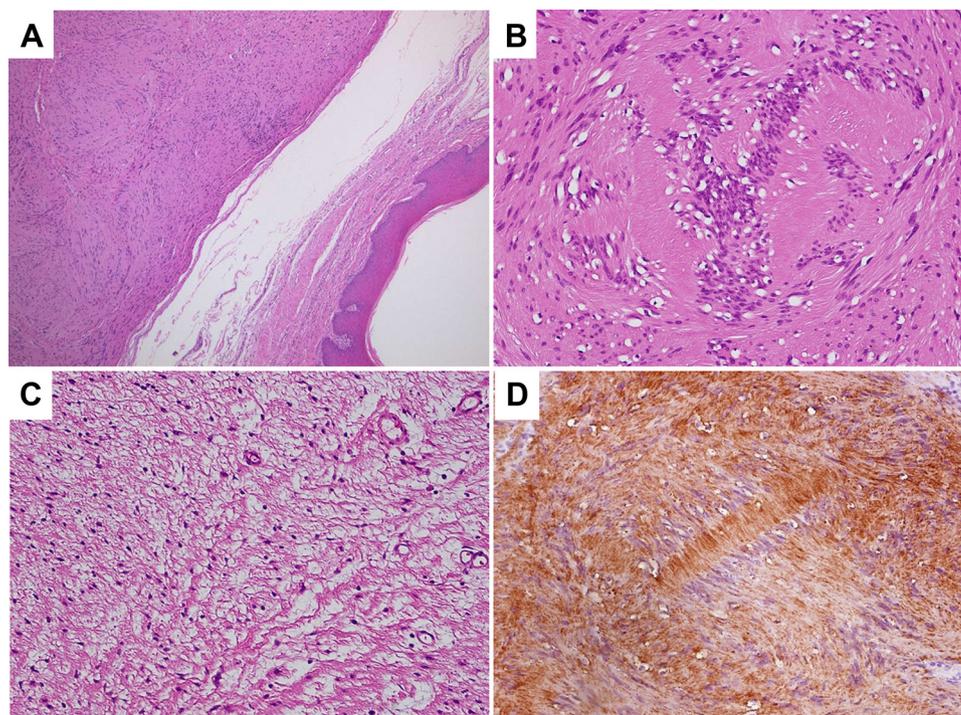


Fig. 6. Histopathological images. The tumor of the squamous epithelium with relatively clear boundary and proliferation of the spindle-shaped cells (A: HE staining). Antoni type A pattern consisting of palisading spindle-shaped cells (B: HE staining). Antoni type B pattern containing a myxoid stroma (C: HE staining). Immunohistochemistry positive for S-100 protein (D: immunostaining). Bars = 100 μm



Fig. 7. Radiographs of 1 year after surgery.

Open bite remains in the anterior teeth, but occlusion is attained in the molars (panoramic radiograph; A). The inclination of the anterior teeth has improved, and the lips are closed (lateral roentgenographic cephalogram; B).

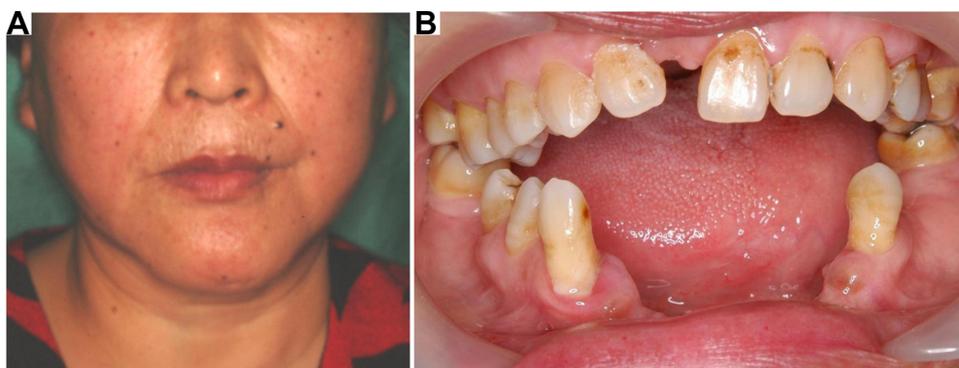


Fig. 8. Photographs of the facial appearance and oral cavity of 9 years after surgery (A: frontal view, B: lateral view, C: oral cavity).

or Neurilemmoma was named based on the findings of collective proliferation of Schwann cells and the basement membrane structure between the interstitium and cells [1]. The etiology of schwannoma is unknown. There is no significant difference in incidence by gender. Schwannomas may occur at any age but more frequently occur between the ages of 10 and 40 [2–5]. Although schwannomas develop in any part of the body with nerves, approximately 25–45% of all schwannomas occur in the head and neck [3]. Of these, approximately 1% occur in the oral cavity, and the most common intraoral sites are the tongue, followed by the palate and oral mucosa [2,5]. In the tongue, tumors are more frequently located in the anterior one-third of the tongue compared with the posterior two-thirds of the tongue [6]. In our case, the tumor had slowly grown over 30 years and reached the center of the tongue. Although the initial site of the tumor is unknown, it was probably located in the left anterior one-third of the tongue. Schwannomas of the tongue present different symptoms depending on the location and size of the tumor [2]. Schwannomas are often left untreated because of their painless nature. The average size of schwannomas of the tongue is approximately 20 mm, and larger tumors of 30 mm or more usually present some symptoms [2,5,6]. The largest size of schwannoma of the tongue in previous reports is 85 mm [7]. Since the tumor in our case also was less symptomatic in the early stage, it reached 95 mm in the maximum diameter. This tumor is the largest schwannoma of the tongue ever reported. When schwannomas are located in the base of the tongue, symptoms such as sore throat, snoring, dysphagia, dysarthria, sleep apnea, dysphonia, and voice change are frequently present [5–7]. On the other hand, when schwannomas are located in the anterior side of the tongue, these symptoms are less frequently present. However, in our case, the mass was so giant that labial inclination of the upper and lower anterior teeth and difficulty in closing the mouth, dysarthria were observed.

For a slow-growing tongue mass that has been present for a long period of time, soft tissue neoplasms and reactive lesions need to be considered. In addition to schwannomas, the differential diagnoses

should include granular cell tumors, leiomyomas, rhabdomyomas, hemangiomas, lymphangiomas, lipomas, pyogenic granulomas, irritation fibromas, neurofibroma and benign salivary gland tumors [8–10]. MRI examination is useful in these differential diagnoses for a tongue mass [2,5]. However, in addition to the imaging examination, the histological examination is required for a definitive diagnosis of schwannoma. Histopathologically, schwannoma consists of two types of characteristic patterns: Antoni type A and Antoni type B. Antoni type A shows dense tumor cells with spindle-shaped nuclei in a palisading arrangement and forms Verocay bodies. Antoni type B is characterized by sparse tumor cells in a reticulated arrangement and vacuolar degeneration of interstitial tissue. It is not uncommon that both types coexist in schwannomas [11–13]. The immunohistochemical staining is also essential for the pathological diagnosis of schwannoma. Most tumor cells of schwannoma show a positive reaction to S-100 protein [14,15].

Complete surgical resection is the standard treatment for schwannomas [13], including schwannomas of the tongue [2]. For the surgery, either an intraoral approach or extraoral approach is selected depending on the location and size of the tumor. In our case, the tumor was giant, but it was located where the intraoral approach could be performed by retracting the tongue. In addition, preoperative airway assessment is important because the location and size of the tumor may be associated with the risk of airway obstruction at the time of inducing general anesthesia [16]. Nasotracheal intubation was performed using the awake fiberoptic technique in our case since mask ventilation and orotracheal intubation were difficult to be performed due to the giant size and the anterior location of the tumor.

Recurrence has never been reported after the complete resection of schwannoma of the tongue [2]. Although malignant transformation of schwannoma is rare [2,16,17], the long-term follow-up has been ongoing.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Conflict of interest

Authors declare no conflict of interest.

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