Large Palatal Pleomorphic Adenoma Complicated by Dysphagia: A Case Report

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A 60-year-old Japanese woman presented with a palatal pleomorphic adenoma measuring 5 cm in size. In addition to impairments during the oral preparatory and oral transport phases, dysphagia with nasopharyngeal closure disorder was observed in the pharyngeal phase. After resection of the tumor, dysphagia resolved, and the patient was immediately able to eat a standard meal.
A videofluoroscopic swallowing study confirmed improvement in the movement of the soft palate compared with the pre-operative condition.

Key words: palatal pleomorphic adenoma, dysphagia, VFSS(Videofluoroscopic Swallowing Study)

INTRODUCTION

Pleomorphic adenoma is the most common tumor of the salivary glands. Among the major salivary gland, it frequently arises in the parotid gland, and among the minor salivary gland, in the palatal gland. However, a pleomorphic adenoma of the palate measuring 5 cm or larger is rare. We performed surgical treatment in a patient with a very large pleomorphic adenoma in the palate who had dysphagia accompanied by nasopharyngeal closure dysfunction during the pharyngeal phase, in addition to impairments during the oral preparatory and transport phases. After surgery, the patient's swallowing function improved rapidly, and the improvement was maintained without recurrence. Resection of the tumor and subsequent improvement of the movement of the soft palate, as shown by a videofluoroscopic swallowing study, helped restore swallowing functions. Here, we describe the case and present the results of the postsurgical videofluoroscopic swallowing study.

CASE REPORT

A 60-year-old Japanese woman presented to our hospital in 2016 because of difficulty in swallowing, speaking, and breathing ascribed to a large mass in the oral cavity. Although the patient had been aware of a mass lesion in her oral cavity for 3 years, she did not seek medical treatment because it was not painful. However, the tumor gradually grew larger, and the patient found it difficult to swallow. Although this patient had no history of functional dysphagia, her past history included tonsillectomy during childhood. On examination, her face was symmetrical, and she showed reversed occlusion. Inside the oral cavity, a semi-spherical tumor measuring 5 cm with well-defined margins occupied the hard palate and extended slightly to the left soft palate (Fig. 1). There was no mucosal erosion or ulceration. The tumor was firm, and there was no pain on palpation. No open nasality was observed at the time of vocalization. The elevation of the uvula was poor, and the uvula was displaced to the healthy side.

Orthopantomography, CT, and MRI were performed. Orthopantomography did not reveal anything other than periodontal disease. CT did not show evidence of bone absorption related to the tumor. On MRI, the tumor measured 57 × 33 × 36 mm and occupied the region from the left hard palate to the soft palate, compressing the upper pharynx. The lesion showed a clear margin with the surrounding tissues and contained heterogeneous high and low signal intensity areas on T2-weighted images. A low intensity capsule-like structure was seen at the tumor margins (Fig. 2A, B, C). The patient complained that hard food was difficult to masticate and transport from the oral cavity to the pharynx because the tumor hit the tongue, and non-viscous fluids sometimes flowed back toward the nasal passages. The preoperative FOIS (functional oral intake scale) was Level 5 (Table 1) [1].

Histopathological diagnosis of pleomorphic adenoma was made by open biopsy. Tumor resection was performed under general anesthesia. The incision line was set at a margin of 5 mm from the tumor to avoid

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Fig. 1 Oral cavity photo at the initial examination. A semi-spherical tumor is present on the hard palate and extends to the soft palate.

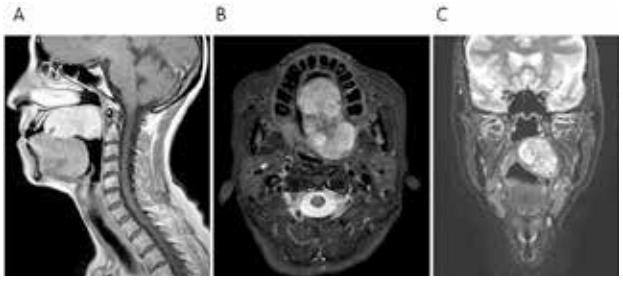

Fig. 2 MRI findings
A: Sagittal T1-W Gd image: The tumor measures 57 × 33 × 36mm.
B: Axial T2-W image. The tumor lesion shows heterogeneous high and low signal intensity areas. A low intensity capsular structure is present at the margins.
C: Coronal STIR image. The tumor compresses the upper pharynx.

Table 1 Functional Oral Intake Scale

Level 1: Nothing by mouth.
Level 2: Tube-dependent with minimal attempts at food or liquid.
Level 3: Tube-dependent with consistent oral intake of food or liquid.
Level 4: Total oral diet of a single consistency.
Level 5: Total oral diet with multiple consistencies, but requiring special preparation or compensations.
Level 6: Total oral diet with multiple consistencies without special preparation, but with specific food limitations.
Level 7: Total oral diet with no restrictions.

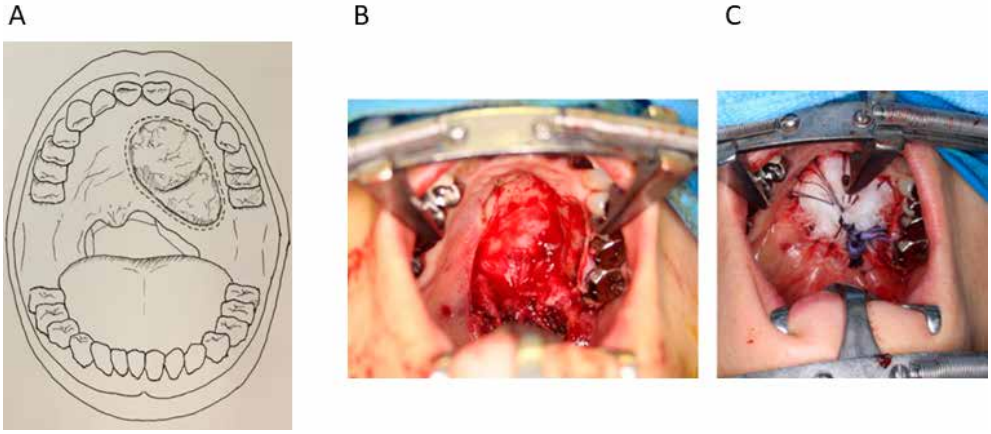

Fig. 3 Surgical findings
A: Oral cavity schema.
B: There were no adhesions between the tumor and surrounding tissues, and dissection was easy. The entire mass was resected.
C: The wound was covered to achieve hemostasis and was protected with a gauze pad.

breaking the tumor capsule on excision. The anterior arch and uvula could be excluded from the excision area (Fig. 3A). After mucosal resection, the tumor was removed from the bone surface at the front. Next, the tumor was manually peeled off, and the entire tumor was removed at once. This enabled preservation of the pharyngeal muscles and avoided damaging the nasal floor mucosa and maxillary sinus mucosa (Fig. 3B). The resected tumor was fully covered by a capsule, and there were no adhesions with surrounding tissues. To achieve hemostasis, the tumor bed was covered with a polyglycolic acid sheet (Neoveil®) and a biosurgical fibrin sealant product (Beriplast®) (Fig. 3C). The resected lesion measured 57 × 33 × 36mm, weighed 38 g, and was spherical (Fig. 4A). Histological examination revealed myxomatous stroma by H-E staining. Ductal

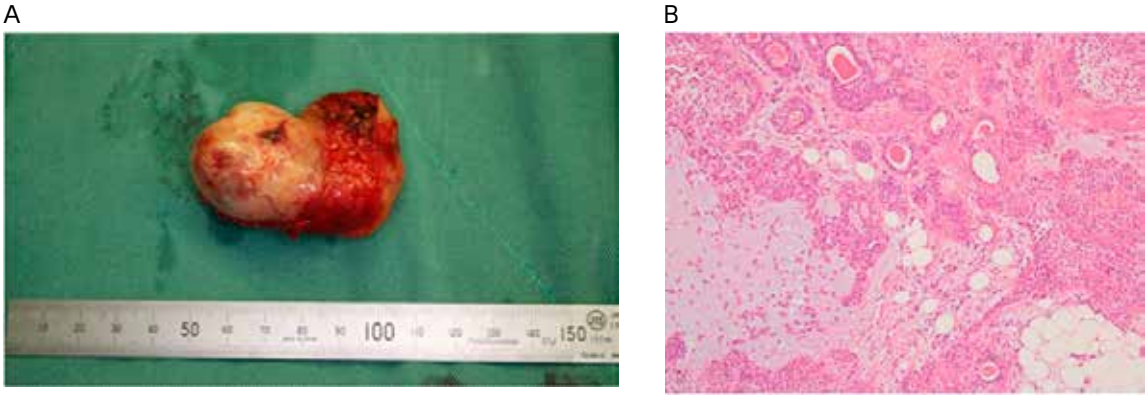

Fig. 4 Extracted specimen
A: The resected tumor weighed 38 grams.
B: Histological findings (H-E staining)

Fig. 5 Postoperative oral cavity findings
Oral cavity photograph at 1 year postoperatively. There is no recurrence of the tumor.

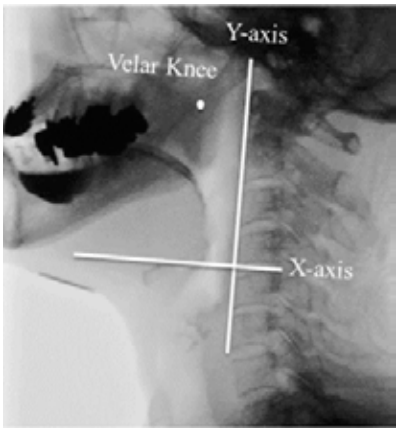

Fig. 6 Lateral videofluoroscopic swallowing image
The X-axis, Y-axis, and velar knee (●) are shown. For the coordinates, the line connecting the anterior lower end of the second cervical vertebra and the anterior lower end of the fourth cervical vertebra was assumed to be the Y axis, and the line perpendicular to the Y axis at the anterior lower end of the fourth cervical vertebra was assumed to be the X axis; the distance was measured between the velar knee and each axis.

structures were present sporadically along with dense proliferation of spindle-shaped myoepithelial tumor cells. Adipocytes were also sporadically present. There was no evidence of malignancy. The histopathological diagnosis was pleomorphic adenoma (Fig. 4B).

Once surgical wound healing was sufficient, a swallowing screening test (revised water drinking test and food test) was performed. Swallowing was judged as possible without the risk of aspiration, and oral intake was started with soft foods from post-operative Day 12. The difficulty of transporting the food bolus from the oral cavity to the pharynx and the complaint of regurgitation of non-viscous liquid toward the nasal cavity had disappeared. The patient was discharged from the hospital on post-operative Day 19 once she had returned to a normal diet level after stepwise feeding and swallowing training (preoperative Level 5 FOIS improved to Level 7 postoperatively). Five years have passed since the operation, and swallowing function is still maintained without a need for functional training related to nasopharyngeal closure, and no obvious relapse has been observed (Fig. 5).

MAIN OUTCOME MEASURES

Videofluoroscopic swallowing study

A videofluoroscopic swallowing study (VFSS) was conducted before surgery and 15 months afterward. While in the sitting position, the patient held 2 ml of barium-containing water in the floor of the mouth and then swallowed it. This procedure was repeated 6 times and each swallowing process was recorded in the lateral view at 30 frames/sec. Before surgery, no residue or aspiration was observed during the oral pharyngeal period, the bulge of the posterior pharyngeal wall was maintained, and regurgitation of barium water into the nasopharynx was not clearly observed. However, there was a decrease in soft palate elevation, which improved after surgery. Therefore, to evaluate the movement of the soft palate in more detail, the soft palate position was analyzed using DIPP-Motion PRO 2D DITECT Corporation, Tokyo, Japan. Posterior and superior velar displacement of the velar knee was

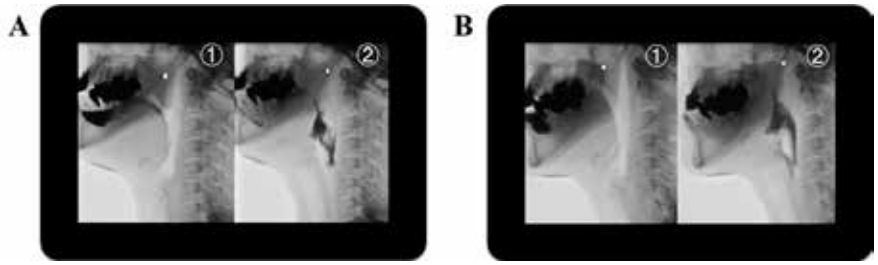

Fig. 7 Position of the velar knee at swallowing
A: Before surgery. B: After surgery.
Velar knee (●); (1) Resting position; (2) Maximum posterosuperior displacement.
The resting position, which was brought closer to the posterior wall of the pharynx because of compression by the tumor, was moved anteriorly by surgical removal of the tumor. Furthermore, the maximum posterior superior orientation of the velar knee during the pharyngeal phase was increased postoperatively.

Table 2 Position of velar knee on the X-axis

	Before surgery	After surgery	P value*
Resting position (mm)	13.6(0.4)	21.3(0.6)	<0.001
Maximum PD (mm)	9.0(0.4)	9.0(0.8)	0.954

PD: posterior displacement. *Paired t-test.

Table 3 Position of velar knee on the Y-axis

	Before surgery	After surgery	P value*
Resting position (mm)	55.6(0.3)	55.8(0.3)	0.391
Maximum SD (mm)	61.4(1.1)	65.0(0.1)	<0.001

SD: superior displacement. *Paired t-test.

measured relative to a Y-axis that connected the anterior lower corners of the second and fourth cervical vertebrae and an X-axis drawn perpendicular to the anterior lower corner of the fourth cervical vertebra (Fig. 6) [2–5].

Data Analysis

Based on analysis of movement in the X-axis direction, the resting velar knee was located at 13.6 mm before surgery and at 21.3 mm after surgery, showing a significant difference. There was no difference in the maximum posterior orientation of the soft palate during the pharyngeal period (Fig. 7, Table 2). In the Y-axis direction, the resting velar knee was located at 55.6 mm before surgery and at 55.8 mm afterward, showing no significant difference. However, the highest superior position of the velar knee was at 61.4 mm before surgery and at 65.0 mm afterward, demonstrating a significant difference (Fig. 7, Table 3).

Statistical Analysis

Normality of the data distribution was confirmed by the Shapiro-Wilk test, and the paired *t* test was used to compare velar displacement before and after surgery. All analyses were performed with the Statistical Package for the Social Sciences (SPSS) version 20.0 (IBM, Amonk, NY), and a p value of less than 0.001 was considered to indicate statistical significance.

DISCUSSION

Pleomorphic adenoma has a high incidence among salivary gland tumors and frequently arises from the parotid gland or the palatal glands [6]. We performed a literature search pleomorphic adenomas arising from the palate like the tumor in the present patient and identified only 3 other cases since 1970 in which the maximum tumor diameter exceeded 50 mm [7–9]. Pleomorphic adenoma generally grows slowly and is not associated with ulceration or pain. Accordingly, symptoms are limited unless there is functional impairment, and it is often overlooked for a number of years, so the majority of patients have a long disease history. However, the incidence of cancer arising in pleomorphic adenoma is reported to increase over time [10]. Malignancy is reported in only 1.5% of pleomorphic adenomas within 5 years after detection, but the rate increases to 9.5% at 15 years or longer [11, 12]. Relapse is generally considered to be common within a few years after surgery, but there is a report of relapse after more than 20 years [13]. Patients with relapse should be assessed carefully because Phillips, *et al.* reported cancer in 7.1% of 126 patients with relapsing pleomorphic adenoma [14]. Therefore, complete resection of pleomorphic adenoma is always required.

For management of the wound after resection of a palatal tumor, a large skin flap may be used to fill the

dead space after removal of a malignant lesion. On the other hand, it has been suggested that a soft skin flap should be used to preserve oral function [15–17]. In patients with benign tumors, it is rare for the incision to extend to the oropharyngeal wall, so reconstruction with a large skin flap is not necessary as long as the posterior mucosa of the soft palate is preserved. The insertion of a flap should be decided based on various risks, including flap necrosis. A method that enhances secondary epithelialization of the wound can be employed or a local skin flap based on the buccal fat pad can be used. If preservation of the palatal mucosa is not possible, it has been suggested that a forearm skin flap with good flexibility may be the most suitable option[18]. Furthermore, a maxillary prosthesis may be used for reconstruction [19]. A previous report described treating the wound by enhancement of secondary epithelialization without a free skin flap, which was similar to our approach, and there was no postoperative functional impairment [7–9].

In the present case, the tumor growing from the hard palate occupied the oral cavity and displaced the soft palate. However, tumor resection and stepwise eating training improved the eating condition from FOIS level 5 to level 7. In other words, the complaint suggesting fluid regurgitation to the upper pharynx disappeared immediately after the operation without training for nasopharyngeal insufficiency, and the patient could resume standard eating. These rapid improvements were considered to be attributed to simple tumor resection without skin flap reconstruction.

Preoperative motion assessment of the soft palate by VFSS showed that the resting position was closer to the posterior wall of the pharynx because of compression by the tumor. However, the resting position moved forwards after surgery (X axis direction, 13.6 to 21.3; $p < 0.001$). Because there was no postoperative change in the maximum posterior orientation during the pharyngeal phase, the functional impact was relatively small. The maximum superior orientation of the soft palate during the pharyngeal phase increased after surgery (Y axis direction, 61.4 to 65.0; $p < 0.001$). In general, closure of the nasopharynx is achieved when the soft palate is elevated and the posterior wall of the pharynx protrudes. In the present case, VFSS showed no abnormal findings of protrusion of the posterior wall of the pharynx either before or after the operation. Accordingly, the reason why the patient's complaint suggesting incomplete closure of the nasopharynx during swallowing disappeared after the operation was considered to be an improvement of the superior movement of the soft palate during the pharyngeal phase after tumor resection. However, in cases without visible structural changes, a more detailed assessment of the oral and pharyngeal phases may be needed to elucidate the mechanisms underlying a preoperative lack of nasopharynx closure. For example, if pre-operative regurgitation does not always occur and viscus fluid does not regurgitate, the timing of nasopharynx closure may be abnormal.

After the operation, muscle spindle functions may be affected because the tensor palati muscle may remain extended for a longer period, even after removal of the tumor. Therefore, several parameters need to be considered, such as the preoperative amount and duration of compression of the soft palate by the tumor and the food passage time (assessed by VFSS).

Finally, according to our literature search, case reports of pre-operative dysphagia due to a benign tumor in the oral cavity are rare. Furthermore, no report describes VFSS assessment of swallowing functions before and after surgical removal of the tumor.

CONFLICT OF INTEREST

There is no conflict of interest with respect to this manuscript.

REFERENCES

1) Crary MA, Carnaby Mann GD, Groher ME. Initial psychometric assessment of a functional oral intake scale for dysphagia in stroke patients. Arch Phys Med Rehabil 2005; 86: 1516–20.
2) Perry JL, Bae Y, Kuehn DP. Effect of posture on deglutitive biomechanics in healthy individuals. Dysphagia 2012; 27: 70–80.
3) Logemann JA, Pauloski BR, Rademaker AW, Colangelo LA, Kahrilas PJ, Smith CH. Temporal and biomechanical characteristics of oropharyngeal swallow in younger and older men. J Speech Lang Hear Res 2000; 43: 1264–74.
4) Steele CM, Bailey GL, Chau T, Molfenter SM, Oshalla M, Waito AA, *et al*. The relationship between hyoid and laryngeal displacement and swallowing impairment. Clin Otolaryngol 2011; 36: 30–6.
5) Kim Y, McCullough GH. Maximal hyoid excursion in poststroke patients. Dysphagia 2010; 25: 20–5.
6) Gnepp DR. Malignant mixed tumors of the salivary glands: a review. Pathol Annu 1993; 28 Pt 1: 279–328.
7) Lazow SK, Colacicco L, Berger J, Gold B. An unusually large pleomorphic adenoma of the soft palate. Oral Surg Oral Med Oral Pathol 1984; 58: 386.
8) Yilmaz AD, Unlü E, Orbay H, Sensöz O. Giant pleomorphic adenoma of soft palate leading to obstruction of the nasopharyngeal port. J Craniofac Surg 2006; 17: 1001–4.
9) Graham AJ, McLoughlin P. Interesting Case: Maxillary swing approach for resection of a large pleomorphic adenoma arising in the soft palate. Br J Oral Maxillofac Surg 2006; 44: 307.
10) Thompson L. World Health Organization classification of tumours: pathology and genetics of head and neck tumours. Ear Nose Throat J 2006; 85: 74.
11) Eneroth CM, Zetterberg A. Malignancy in pleomorphic adenoma: A clinical and microspectrophotometric study. Acta Otolaryngol 1974; 77: 426–32.
12) Seifert G, Sobin LH. The World Health Organization's Histological Classification of Salivary Gland Tumors. A commentary on the second edition. Cancer 1992; 15: 70: 379–85.
13) Akiba J, Harada H, Kawahara A, Todoroki K, Nagata S, Yano H. A case of metastasizing pleomorphic adenoma in the maxillary bone appearing twenty years after initial resection of pleomorphic adenoma of the hard palate. Pathol Int 2013; 63: 463–8.
14) Phillips PP, Olsen KD: Recurrent pleomorphic adenoma of the parotid gland: report of 126 cases and a review of the literature. Ann Otol Rhinol Laryngol 1995; 104: 100–4.
15) Gilas T, Sako K, Razack MS, Bakamjian V Y, Shedd DP, Calamel PM. Major head and neck reconstruction using the deltopectoral flap. A 20 year experience. Am J Surg 1986; 152: 430–4.
16) Biller HF, Ogura JH, Brownson RJ. The forehead flap. technique and complication. Arch Otolaryngol 1973; 97: 316–8.
17) Lacombe V, Blackwell KE. Radial forearm free flap for soft palate reconstruction. Arch Facial Plast Surg 1999; 1: 130–2.
18) McCombe D, Lyons B, Winkler R, Morrison W. Speech and swallowing following radial forearm flap reconstruction of major soft palate defects. Br J Plast Surg 2005; 58: 306–11.
19) Brauner E, Jamshir S, Guarino G, Ciolfi A, Valentini V, Pompa G, *et al*. Pleomorphic adenoma rehabilitative treatment in growing up patient: a 20-years follow-up. Eur Rev Med Pharmacol Sci 2016; 20: 3523–7.