

Transoral endoscopic surgical approach to venous malformation of the parapharyngeal space

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Abstract

We encountered a rare case of venous malformation located in the parapharyngeal space. The 65-year-old female patient did not have any symptoms, and the malformation was discovered incidentally during a clinical survey. Examination of the oral cavity revealed a mass in the left soft palate. Magnetic resonance imaging showed a well-defined mass in the left parapharyngeal space. Fine needle aspiration cytology suggested no malignancy. Four years after the first visit, she underwent surgery for diagnosis and treatment. We safely removed the mass with a rigid videoendoscope trans-orally. No postoperative complications arose, and she was discharged 7 days after the operation. Histopathological examination identified cavernous hemangioma. Venous malformation (cavernous hemangioma) of the parapharyngeal space is very rare, and few cases of removal under a transoral approach using a rigid endoscope with a flexible tip have been reported. This approach is safe and can be recommended for selected tumors of the parapharyngeal space.

Keywords

Venous malformation, cavernous hemangioma, parapharyngeal space, transoral approach, flexible endoscope

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Introduction

The parapharyngeal space (PPS) is a space shaped like an inverted triangle, located lateral to the upper pharynx and medial to the mandible, extending from the skull base superiorly to the hyoid bone inferiorly. Tumors of the PPS are rare, accounting for 0.5%–1.5% of all head and neck tumors.^{1,2} Furthermore, venous malformations (VMs) of the PPS are uncommon, accounting for $\leq 2\%$ of all PPS tumors.^{1,3} We present a case of a female patient with a VM of the PPS, which was resected using a fully transoral endoscopic surgical approach.

Case presentation

A 65-year-old woman with breast cancer was referred to our hospital after incidental identification of an oropharyngeal mass during a clinical survey. She was asymptomatic. Examination of the oral cavity revealed a mass in the left soft palate. Nasopharyngoscopic examination showed prominence in the left pharyngeal wall near the pharyngeal orifice of Eustachian tube (Figure 1). Magnetic resonance imaging (MRI) demonstrated a well-defined mass in the left PPS,

measuring 20 mm \times 18 mm \times 28 mm. The tensor veli palatini muscle was displaced medially, while the medial pterygoid muscle was displaced laterally by the mass. The mass appeared to be separate from the deep lobe of the parotid gland. T1-weighted imaging showed the lesion with intermediate signal characteristics. Gd-enhanced T1-weighted imaging indicated high signal intensity from the center to the outside. Signal intensity was mostly high on T2-weighted imaging, but with a few areas of low signal intensity (Figure 2). Fine needle aspiration cytology revealed only components in the blood serum and did not suggest malignancy. She had been followed up with annual MRI for 4 years. Despite remaining asymptomatic, she was concerned that the mass had not shrunk remarkably. Thus, she decided to have surgery for both diagnosis and treatment.

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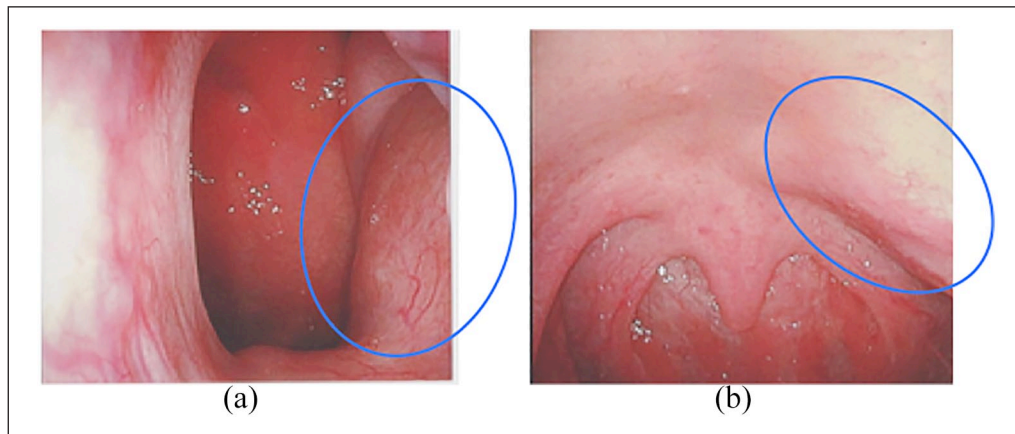


Figure 1. Nasopharyngoscopic examination of the oropharynx and direct examination of the oral cavity revealed a mass in the left soft palate. (a) Swelling of the pharyngeal orifice of Eustachian tube and (b) a mass in the left soft palate.

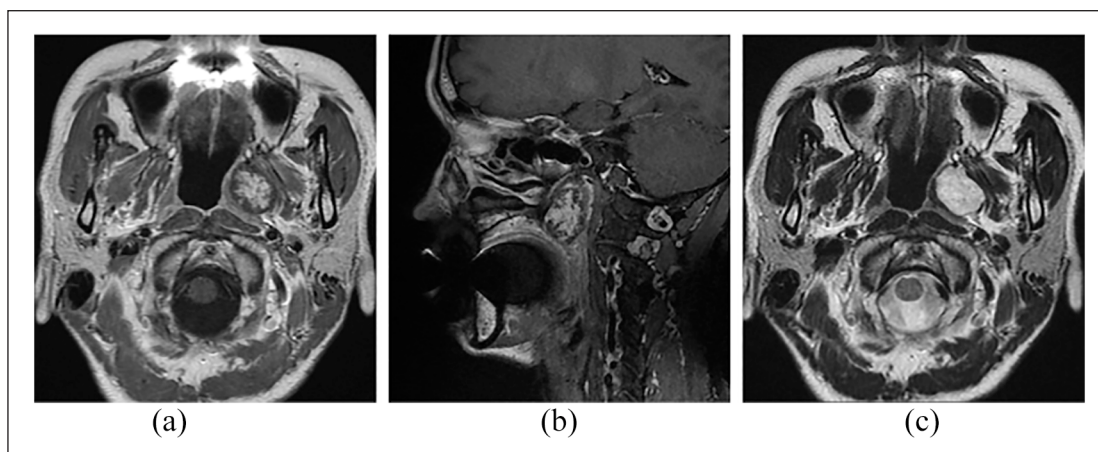


Figure 2. MRI before the operation showed the well-defined mass in the left PPS: (a) axial Gd-enhanced T1-weighted, (b) sagittal Gd-enhanced T1-weighted, and (c) axial T2-weighted.

Surgery

The operation was performed under general anesthesia with nasal intubation. The surgical view was obtained using a Crowe-Davis retractor. An assistant maintained a view of the pharynx using a deflectable-tip ENDOEYE (OLYMPUS LTF-S190-5, Tokyo, Japan) (Figure 3). First of all, a vertical incision was made in the mucosa of the soft palate and then the constrictor muscle of pharynx was cut to expose the surface of the tumor capsule. Adipose tissue of the PPS and medial pterygoid muscle were easily identified lateral to the tumor. The rigid endoscope with the flexible tip could provide a clear surgical field that allows for secure dissection from a deep surgical site such as behind the hard palate. The tumor was shrinking gradually by ligation of the feeding vessels. Finally, the tumor was successfully removed with little bleeding. The resected specimen was soft and spongy (Figure 4).

Oral intake was initiated on postoperative day 3. The patient was discharged on postoperative day 7. No postoperative complications were encountered. Histopathological

examination demonstrated VM (Figure 4). At the 6-month follow-up, the patient showed no evidence of residual tumor.

Discussion

Around 80% of PPS tumors are benign, predominantly in the form of salivary gland tumors followed by neurilemmomas.¹ Kale et al.⁴ first described a female case of PPS cavernous hemangioma in 2006. “Cavernous hemangioma” has been used as a colloquial term for VMs. The Current International Society for the Study of Vascular Anomalies Classification System groups vascular anomalies into two categories: tumors and malformations. VM is the most common vascular malformation. This entity has been recognized as distinct from hemangioma, with the latter considered as a true neoplasm.⁵ A VM is a slow-flow blood storage lesion with a venous cavity that has become cystic and spongy because of the extensive capillary component resulting from hypoplasia of vascular endothelial cells in the course of vasculogenesis during embryogenesis. More than 90% of cases are sporadic,

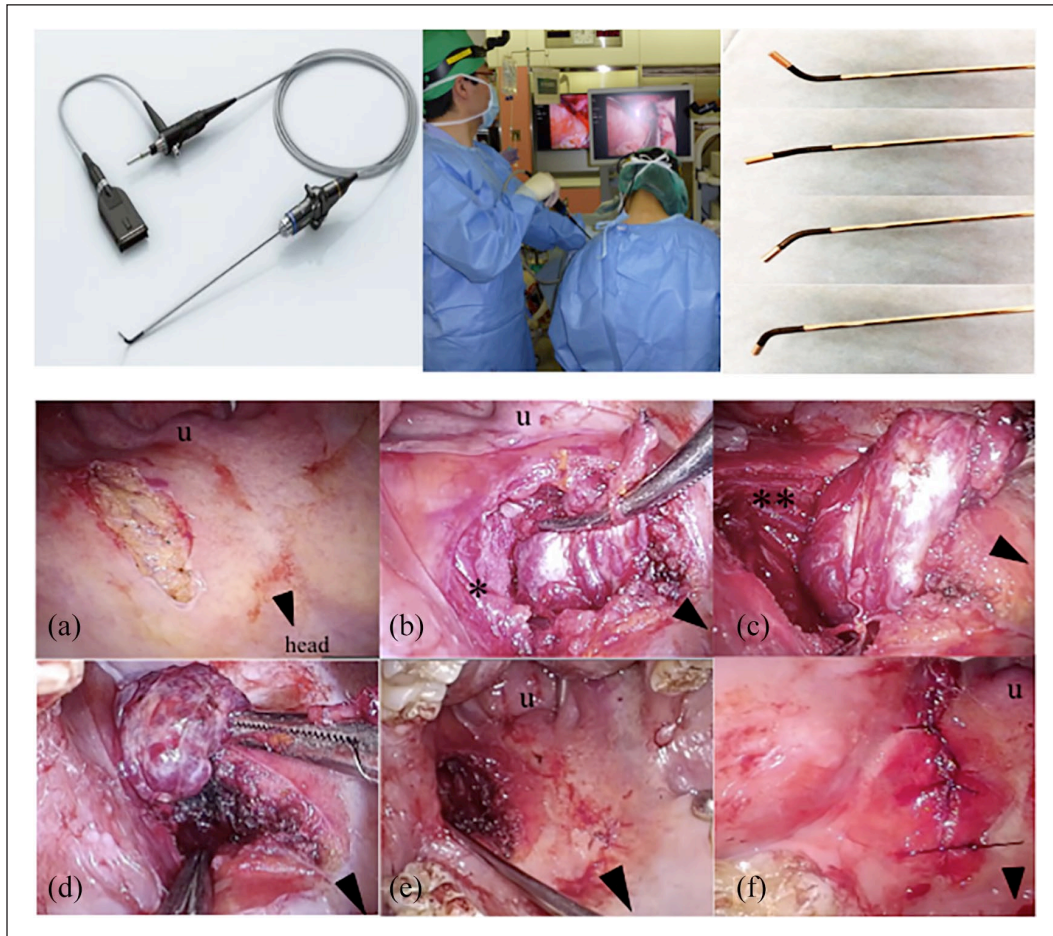


Figure 3. Surgical view. We could clearly visualize the tumor using a flexible-tipped rigid endoscope (deflectable-tip ENDOYEYE; OLYMPUS LTF-S190-5, Tokyo, Japan). (a) A vertical mucosal incision was made in the soft palate; (b) the constrictor muscle of the pharynx was cut, and the surface of the tumor capsule was exposed; (c) the tumor was separated from the medial pterygoid muscle and the tissue behind the hard palate; (d) the tumor was completely excised; (e) the wound after the excision; and (f) the wound was sutured. u: uvula.

*The medial pterygoid muscle.

**The tensor veli palatini muscle.

with some inherited cases.⁶ Histopathological examinations show extensive venous vasculature in the connective tissue, with thin vascular walls. On MRI, T1-weighted imaging shows intermediate or low signal intensity. On T2-weighted imaging, signal intensity is high. Gd-enhanced MRI shows slow tumor staining, which is useful for differentiation from lymphangioma. MRI findings in our case were consistent with those of the literature. Diagnosis of hemangioma by fine needle cytology is considered difficult, due to the excessively bloody specimen.

The choice of the surgical approach for PPS tumor depends on tumor size, location, and histopathology. The main goals of this surgery are to obtain good exposure and direct access, to protect major vessels, and to preserve physiological functions. Four main surgical approaches have been used: transcervical, transparotid, mandibular swing, and transoral.^{7,8} The transcervical approach has been conventionally chosen through the course of long

years of experience. The transoral approach was previously not been adopted because of the disadvantages of poor exposure, inability to control bleeding, nerve injury, and incomplete removal. The transoral approach initially described by Ehrlich in 1950 is indicated for small, non-vascular tumors.⁹ Recently, the clinical situation has changed with the development of endoscopy and surgical robotics. Therefore, several case reports have examined the transoral approach to PPS tumors. Meng et al.¹⁰ stated that the transoral endoscopic approach is suitable for tumors medial or anteromedial to the internal carotid artery (ICA), but not for tumors with no clear boundary or features of invasion to the ICA or cranial nerves IX-XII. Our case fulfilled the recommended surgical indications. Chen et al.¹¹ conducted a literature review of tumors in the PPS using an endoscopically assisted transoral approach. They described a clear surgical view is indispensable to avoid blind manipulation in deep areas of the pharynx.

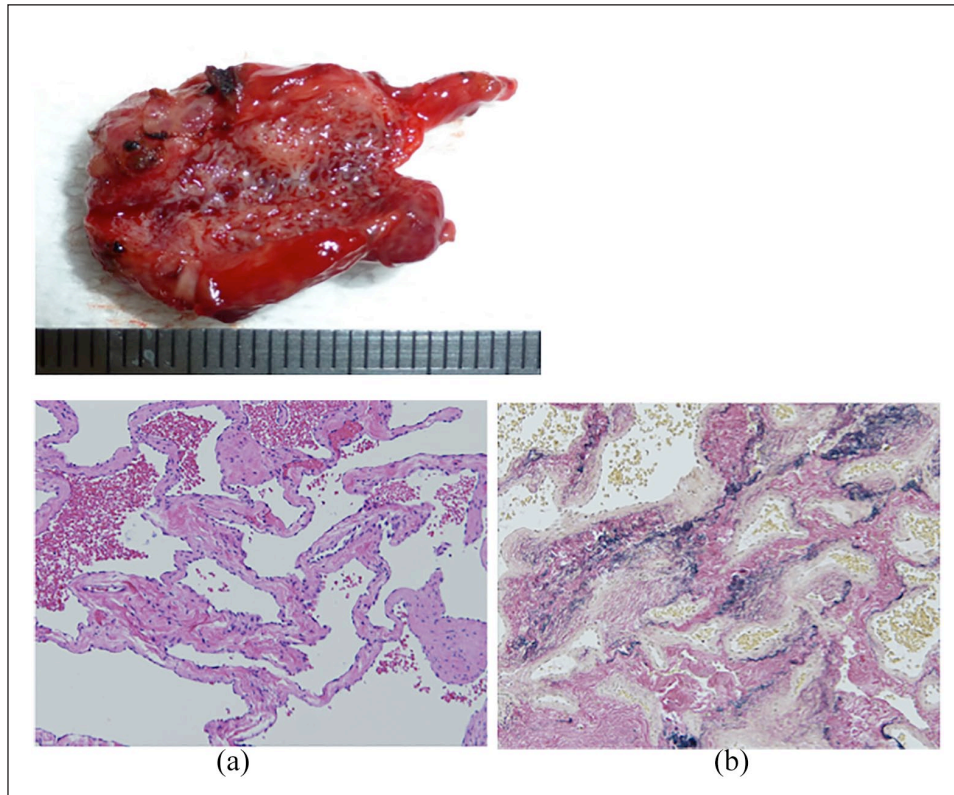


Figure 4. The resected specimen was soft and elastic. Results from histopathological examination: cavernous hemangioma. (a) Hematoxylin and eosin (HE) staining. The extended venous cavity is surrounded by a single layer of vascular endothelial cells. (b) Elastica van Gieson (EVG) staining: elastic fibers stain black-purple, indicating blood vessels.

Regarding resection for parapharyngeal VM at PPS, several recent reports have described transoral fully robotic dissection.^{12,13} However, to the best of our knowledge, there are no reports of transoral VM resection with easy-to-use surgical instruments. Besides, because the tumor is benign and shrinks by ligation of the feeding vessels which facilitated to remove the mass during the operation, from this experience oral resection without robotic surgery may be required depending on the circumstances of the facility; thus, this case can represent a rare description of VM in the PPS. We applied the transoral videolaryngoscopic surgery (TOVS) method to this case. TOVS has been developed for laryngopharyngeal lesions by Shiotani et al.¹⁴ In comparison with transoral robotic surgery (TORS), TOVS is easy for otolaryngologists to perform without special training because it is an extension of tonsillectomy or laryngomicrosurgery. TOVS can also provide advantage similar to those of robotic surgery, such as wide field of view and working space using the ENDOEYE FLEX rigid endoscope with a flexible tip manipulated by an assistant.^{15,16} The high-definition camera provides a clear view, so we could easily identify the tumor margin, peripheral vessels, and nerves. The ENDOEYE FLEX allows viewing behind the hard palate because the tip of the endoscope can be flexed by 100°, relative to the initial position. It was unnecessary to exchange the endoscope tip due to the ability to change the angle of the tip freely during

surgery. In conclusion, the biggest advantage is that this technique allows safe manipulation under clear visualization without extravagant equipment.

Conclusion

We encountered a rare case of VM of the PPS. We could visualize and safely remove the tumor using the rigid endoscope with a flexible tip. The tumor was shrinking gradually which facilitated to remove the mass during the operation.

This approach is safe, less invasive, and can be recommended for PPS tumors in carefully selected cases, especially for VM.

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Declaration of conflicting interests

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Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

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Informed consent

Written informed consent was obtained from the patient for their anonymized information to be published in this article.

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