

CASE REPORT

Endoscopic-assisted resection of pleomorphic adenoma in the accessory parotid gland

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Abstract : Background : An accessory parotid gland (APG) is a common anatomical structure that occurs in 10%–56% of individuals. Pleomorphic adenomas are the most common benign tumors of the APG, and their ideal treatment is surgical excision, although there is a risk for aesthetic disorders and facial nerve damage due to the site of origin. Moreover, despite being benign, these tumors are known to recur. Therefore, it is necessary to achieve both reliable excision and avoidance of facial nerve damage. **Case presentation :** We report a case of a 49-year-old Japanese man with a mass in his left cheek. The lesion was diagnosed as a benign salivary gland tumor derived from the APG by computed tomography imaging, magnetic resonance imaging and fine needle aspiration cytology. We resected the tumor using modified high submandibular incision under the endoscopic-assisted field of view. **Discussion and Conclusions :** The tumor was less invasive and reliably resected using an endoscope. In surgical treatment, the endoscopic-assisted technique is very useful to achieve complete tumor resection and prevent relapse while avoiding serious complications due to surgical procedures. *J. Med. Invest.* 68 : 376-380, August, 2021

Keywords : Endoscopy, Pleomorphic adenoma, Accessory parotid gland, Minimally invasive surgery

BACKGROUND

An accessory parotid gland (APG) is a common anatomical structure that occurs in 10.2%–56% of individuals (1, 2). The type of APG lesions is similar to that of parotid gland lesions, and 1%–8% of all parotid gland lesions are derived from the APG (3). Tumors of the parotid gland and APG can be differentiated on the basis of whether continuity can be detected between the tumor and the parotid gland (4).

Pleomorphic adenomas are the most common benign salivary gland tumors, especially in the parotid gland, and their ideal treatment is surgical excision. Extracapsular dissection using the external preauricular incision approach is sufficient to treat the majority of pleomorphic adenomas in APG, and the conventional transcuteaneous excision approach is a well-established surgical technique for complete resection of these lesions (5). However, this method has the major complication of leaving large scars on the facial area.

In recent years, various endoscopic-assisted surgeries have been reported in the field of head and neck surgery (6). A safe surgery requires a large incision, but postoperative scarring becomes serious. Small incisions made to reduce scarring can compromise the safety of surgery. Endoscopic-assisted surgery has become widespread to solve this paradox. Maintaining a good surgical field of view is important to avoid damage to

commonly important anatomical structures such as the nerve and blood vessels. This surgical procedure approached from a small incision has expanded the surgical field of view through the magnifying field of view effect of the endoscope and also strengthened minimally invasive and safe surgery (7).

We report a case study in which reliable and minimally invasive treatment for a pleomorphic adenoma in the APG was performed via endoscopic-assisted surgery.

CASE PRESENTATION

A 49-year-old Japanese man had a history of a painless mass in his left cheek, which had been slowly growing in size over the past 6 months. He was referred to our department at Kagawa Prefectural Central Hospital in 2019 by his local dentist. He had no medical or family history and was healthy.

Extraoral examination revealed a round, painless mass measuring 30 × 25 mm in diameter with no pulsation located anterior to the left masseter muscle and directly inferior to the zygomatic buttress. The mass was not tender and did not change in size during mandibular movements or the Valsalva maneuver. The overlying facial skin was normal, and no adenopathy was noticed. (Fig. 1. a, b) The patient had no history of trauma and surgery in that region. Intraoral examination showed good

List of abbreviations
APG, Accessory parotid gland
FNA, Fine needle aspiration

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saliva flow with firm palpation of the mass in question. It was not possible to palpate the mass from the oral cavity.

A computed tomography scan disclosed a well-demarcated round mass lesion within the left APG. (Fig. 1. c, d) Magnetic resonance imaging revealed a lobulated ovoid mass measuring 31 × 26 × 25 mm with well-defined margin in the left APG. (Fig. 1. e) The lesion showed isointense signals on T1-weighted sequences and a high signal on T2-weighted sequences. (Fig. 1. f) Sample collection using fine needle aspiration (FNA) was appropriate and suggested a benign tumor of the salivary gland. On the basis of the findings of imaging and tissue biopsy, the preoperative diagnosis was a pleomorphic adenoma derived from the APG.

The patient intended to undergo surgical resection in the most aesthetic manner possible. Considering the benign nature of the tumor, we decided that the high submandibular approach, which is often used in oral and maxillofacial surgery in recent years for diseases of the temporomandibular joint, should be combined with endoscopic resection. Surgical excision was performed through an endoscopic-assisted technique under general anesthesia. A skin incision measuring approximately 6 cm in length was made along the retromandibular space via the high submandibular approach. The scalp flap was raised, and a mo-

nopolar ablation and right-angled retractors were used to expose the superficial layer of the parotid gland via the masseter muscle, exposing the entire flap. The next step was to identify the connective tissue around the tumor. Detachment was performed from the surrounding tissue to remove the tumor, including the capsule. In addition to the direct field of view, the endoscopic auxiliary field of view was useful. In particular, the part of the tumor in contact with the masseter muscle was suitable for safe and minimally invasive surgery in the magnified surgical field using the endoscope. A 30° angle, 4-mm Karlstorz endoscope with a cold light source was inserted and replaced to a 70° angle as needed. (Fig. 2)

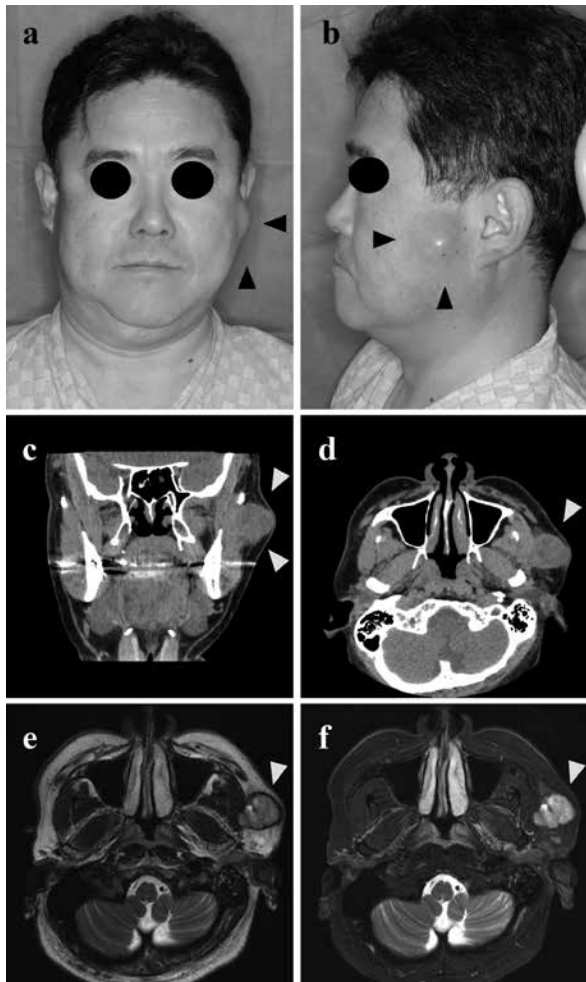


Fig 1. Patient presenting with a swelling of the left mid-cheek area (a, b) Front and side facial photos. (c, d) Coronal and axial images of computed tomography scans showing a tumor outside the left masseter muscle, with no involvement between the tumor and the zygomatic bone. (e) T2-weighted magnetic resonance images showing that the tumor is covered with a thick capsule but partially penetrates the masseter muscle. (f) Fat-suppressed T2-weighted images showing that the tumor is lobulated and internally heterogeneous.

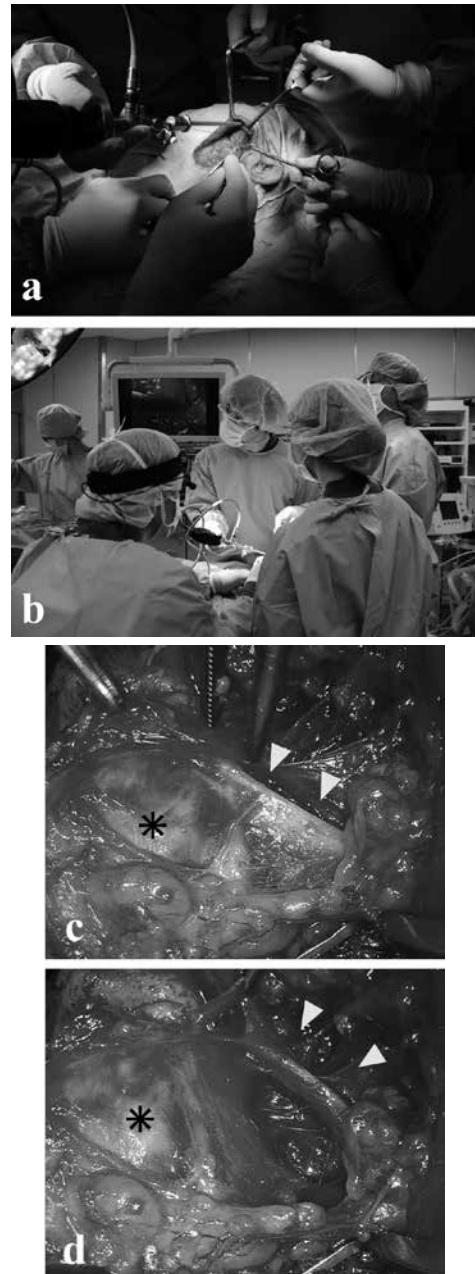


Fig 2. Intraoperative and endoscopic photographs (a) Intraoperative view of the pleomorphic adenoma through a preauricular and high perimandibular approach using an endoscopic-assisted technique. (b) The surgeon and assistants share the same surgical field through the endoscopic monitor. (c, d) The tumor (*) and the buccal branch of the facial nerve (arrowhead) are safely separated in the endoscopic visual field.

Histopathological examination showed proliferation of spindle myoepithelial cells with hyalinizing stroma. (Fig. 3a) In some area, there is ductal structure lined by epithelial cells. (Fig. 3b). The tumor composed of both myoepithelial and epithelial cells, the diagnosis of pleomorphic adenoma was made. The tumor was surrounded by a thick fibrous capsule and contained preexisting salivary gland tissue at some edges. Margin assessment of the capsule revealed an intact tumor capsule with no evidence of tumor penetration. The postoperative clinical course was good, and there were no indications of temporary nerve paralysis. At one year after the operation, the course is good without recurrence, and there are no aesthetic discomforts.

DISCUSSION

APGs are independent of the parotid glands themselves, positioned anterior to them, and superior to the masseter muscle (2, 8). The APG is a common structure, and tumors that occur in the APG are consistent with tumors of the parotid gland. Tumors that should be differentially diagnosed from APG tumors are those developing in the cheek region, including benign tumors such as schwannoma, dermoid cyst, and lipoma, as well as tumors of the lymph node and masseter muscle, Stensen's duct primary tumors, minor salivary gland-derived tumors, and aberrant salivary gland tumors. The histological subtypes of these tumors can be differentiated according to histopathological findings (4). FNA is a commonly performed diagnostic test in the initial evaluation of a parotid mass. The advantage of this technique is that it can be performed in the outpatient setting with minimal recovery time and low risk of complications. However, a potential disadvantage is that it has been associated with variable sensitivity and specificity in distinguishing malignant from benign disease. Moreover, high rates of nondiagnostic aspirations have been reported in the literature (9). FNA of the

parotid gland has moderate sensitivity and high specificity in differentiating malignant from benign disease. Given the high positive likelihood ratio, a positive FNA finding can predict the presence of malignancy with 98%–100% accuracy, depending on the prevalence of the malignancy (10). In our case, sampling via FNA was appropriate and suggested a benign tumor of the salivary gland with no malignant findings. We diagnosed the tumor as a pleomorphic adenoma in the APG based on histological and imaging findings and the frequency of occurrence.

Pleomorphic adenomas account for up to two-thirds of all salivary gland neoplasms, especially in the parotid gland (11). Although this tumor occurs in individuals of all ages, it is most common in the 30–60 years age group and gender differences are more common in female (12). Treatment of this tumor is no longer enucleation in contemporary surgical practice, and partial or total parotidectomy or extracapsular dissection is the operation of choice at initial presentation (13). Pleomorphic adenomas have a low risk of recurrence. After superficial parotidectomy, the recurrence rate may be approximately 2% (14). Tumor-associated factors that may affect recurrence include its size, histopathological subtype, satellite nodules, and inadequacy of encapsulation. Surgery-associated recurrence factors include tumor leakage, infiltrated edges, or both. Recurrent pleomorphic adenomas can pose a treatment challenge because the operation is difficult, the tumor is often multinodular (15), and it can be associated with compromise of the facial nerve. Furthermore, the risk for malignant transformation after recurrence is 3.3% (16). Therefore, reliable excision of the tumor is essential.

The most common conventional approaches for the resection of benign tumors in the APG are a percutaneous S-shaped incision similar to parotid tumor surgery and a face-lift incision. The resulting scars may severely affect the postoperative appearance of the patient (17). Although adequate exposure of the operative field is achieved in traditional approaches, they are subject to several complications, including damage to Stensen's duct, facial

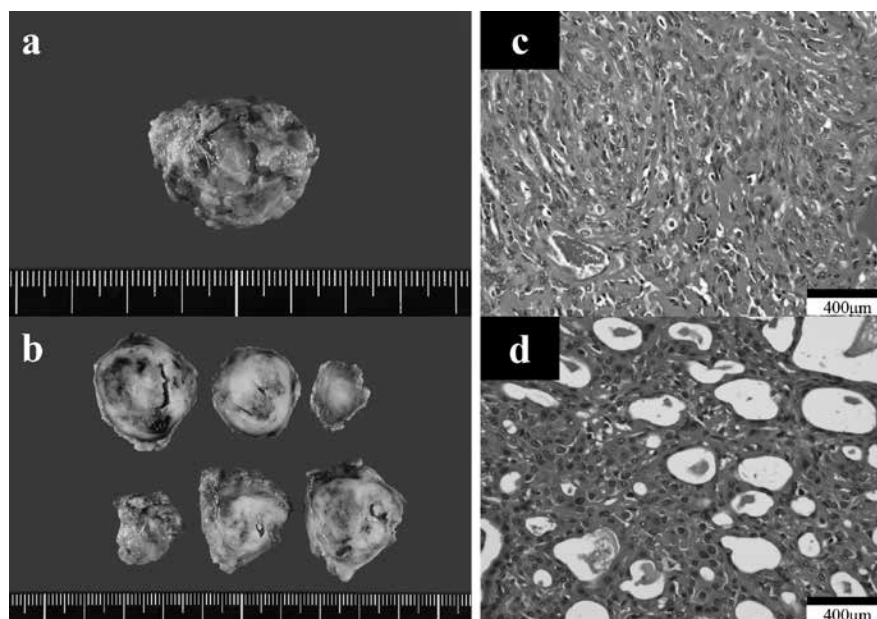


Fig 3. Gross and histological findings

Gross findings of the resected material

(a) The resected tumor was a nodular solid mass measuring $3.2 \times 3.0 \times 2.5$ cm³ in size. (b) The cut surface of the tumor was well defined and white-to-yellowish-brown in color. The capsule and soft tissue around the tumor were also resected. (c) The tumor showed predominately myoepithelial cell proliferation with fibrous matrix (100 \times). (d) In the tumor, proliferation of glandular cells along with myoepithelial components was also observed (100 \times).

nerve injury, skin scars, and salivary fistula. A new surgical method using an endoscope has been proposed to improve these adverse events. Li *et al.* (17) reported a method of removing pleomorphic adenomas and benign lymphoepithelial lesions within the APG under complete endoscopic field of view with an incision along the tragus and two incisions for inserting the endoscope. Zhang *et al.* (18) reported an approach of preauricular incision in the endoscopic-assisted field of view to remove fibromas, hemangiomas, and lymphomas within the APG. A general consensus is that tumors of the parotid or submandibular gland, especially pleomorphic adenomas, should be partially excised around the extracapsular tissue (19). Therefore, for pleomorphic adenoma derived from the parotid gland, high skill is required for endoscopic operation due to the anatomical positional relationship in front of the parotid gland. A modified high submandibular incision is a relatively used approach in the field of oral surgery for treating fractures of the temporomandibular joint process (20). To our knowledge, our case study is the first report to demonstrate that a modified high submandibular approach under endoscopic-assisted visual field may reliably resect the tumor and reduce postoperative complications such as facial nerve injury and scarring.

Endoscopic-assisted surgery can magnify the surgical view and thus enhance the identification of small vessels, nerves, and other anatomically important structures. Maintaining a good surgical view is important to avoid neurovascular damage and injury of important structures. The reported benefits of this method include minimal postoperative scars, improved aesthetic effects, and quicker postoperative recovery (6). In our case also, we were able to shorten the incision line and the buccal branch of the facial nerve could be safely detached from the lesion. Using an endoscope intraoperatively has additional benefits as well. Surgical field confirmation by multiple operators is possible at the same time, and they can communicate smoothly. Endoscopes are also useful for education (21). However, because of the poor operation space, endoscopic-assisted partial resection around the extracapsular tissue requires a highly skilled technique, which prevents its popularization (22). It is important to use endoscopes frequently to train surgeons. Complete endoscopic surgery has also been reported (19), although it is extremely difficult. The immaturity of the assistant causes prolonged surgery time and inadequate surgical visual field expansion. Improving the skills of the first assistant's endoscopic operation is very important for the success of endoscopic surgery. By adding an endoscopic technique to the approach that oral surgeons always use, a safer and less invasive surgery can be performed more easily. The immaturity of the assistant causes prolonged surgery time and inadequate surgical visual field expansion. As our measure, we usually have used endoscopes as a team to create an environment where young assistants can master the operation of endoscopes. Actively incorporating endoscopes into oral surgery is useful not only for patients but also for the future of oral surgery.

CONCLUSIONS

Although pleomorphic adenomas are benign tumors, they require reliable resection to prevent recurrence. Aesthetic impairment is the major problem in the surgical resection of lesions originating from the APG and parotid glands. Endoscopic-assisted surgery is advantageous in terms of both reliable tumor resection and aesthetics.

CONFLICTS OF INTEREST AND SOURCE OF FUNDING

None declared.

PATIENT CONSENT

Written patient consent was obtained to publish clinical photographs.

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