Case Report
Coincidental Tumors in the Main and an Accessory Parotid Gland

Shinichi Asamura,1 Satoshi Nimura,2 Hirohiko Kakizaki,3 Yoshitaka Wada,1 and Noritaka Isogai1

1Department of Plastic and Reconstructive Surgery, Kinki University Faculty of Medicine, 377-2, Ohno-higashi, Osaka sayama, Osaka 589-8511, Japan
2Department of Pathology, Faculty of Medicine, Fukuoka University, 8-19-1, Nanakuma, Jonan-ku, Fukuoka 814-0180, Japan
3Department of Ophthalmology, Aichi Medical University, Nagakute, Aichi 480-1195, Japan

Address correspondence to Shinichi Asamura, asamura@med.kindai.ac.jp

Received 6 November 2012; Accepted 22 March 2013

Abstract
Introduction. We are presenting a rare case of a patient who developed simultaneous tumors in the main parotid gland (MPG) and an accessory parotid gland (APG). The APG tumor was not recognized when the more prominent MPG tumor was excised. We will discuss the clinical features of this case. Case presentation. A 66-year-old woman was referred to our department with a painless right cheek mass. Her past medical history was notable for a right MPG tumor excision at the age of 62. A retrospective review of the original MRI films revealed that a small APG tumor was present prior to the removal of the MPG tumor. Conclusion. Although the coincidental presence of the MPG and APG tumors is quite rare, this case shows the importance of identifying the smaller and potentially more sinister lesion involving the APG when it is present while preparing for the MPG tumor excision.

Keywords accessory parotid gland; main parotid gland; facial nerve; Stensen’s duct; central cheek; malignancy rate

1. Introduction
The accessory parotid gland (APG) is isolated salivary gland tissue, separate from the main parotid gland (MPG), which also drains into the common parotid duct. The presence of an APG is not uncommon, but the tumors originating from APG are relatively rare [1, 2]. We are presenting a rare case of a patient who developed simultaneous tumors in MPG and the APG; the APG tumor was not recognized when the more prominent MPG tumor was excised. We will discuss the clinical features of this case, current management practices, and a review the relevant literature.

2. Case presentation
In June 2011, a 66-year-old woman was referred to our department with a painless right cheek lesion that had grown over the past 3 years. Our initial examination revealed an approximately 2 cm, firm, and non-tender mass, which was located in the center of the right cheek. It was freely mobile; there was no evidence of a sensory deficit or facial nerve palsy. This patient’s MRI demonstrated a well-demarcated 2-cm ovoid soft tissue lesion on the lateral aspect of the right masseter muscle; it did not appear to infiltrate the surrounding tissue or bone. The radiographic diagnosis was a benign pleomorphic adenoma originating from an APG (Figure 1(a)). Of note, this patient had had a basal cell adenoma in her right MPG excised 3 years prior to her presentation to our department (Figure 1(b)).

Under general anesthesia, a facelift incision was made along the scar tissue of the previous incision while taking painstaking care to preserve the zygomatic and buccal branches of the facial nerve. The dissection was advanced onto the plane of the parotid-masseteric fascia. After en bloc dissection of the APG tumor from adjacent tissues, the accessory parotid duct, draining into the main parotid duct, was ligated. The gross appearance of the tumor was similar to a cystic structure, and its content was homogeneous liquid and purple in color (Figure 2(a)).

Figure 1: (a) Preoperative MRI of the second operation. A lesion with clear margin was found in APG (white arrow). (b) Preoperative MRI of the primary operation. A big lesion in MPG (M) and coincidentally a small lesion with clear margin were found in APG (white arrow).
Figure 2: (a) Operative findings. En bloc dissection of the tumor from adjacent tissues, while full attention is paid to avoid the facial nerve injury. The tumor was originated from APG (white arrow). (b) The tumor and APG (dark arrow) location in this case (modified from APG [5]). M: MPG, B: The buccal branch of the facial nerve, S: Stensen’s duct (Partoid duct).

Figure 3: Histological features of the cystic lesion. In APG, a cystic cavity was surrounded with epithelial-lined fibrous tissue. Oncocytic columnar epithelium lined the luminal surface of this cyst.

A microscopic examination revealed that the luminal surface was lined by oncocytic epithelium (Figure 3). The cyst wall was dense fibrous connective tissue. The adjacent salivary gland parenchyma was compressed and atrophic.

The patient was discharged without any complications; there was no impairment of her right facial nerve at her 6-month follow-up evaluation.

3. Discussion
In order to make the accurate diagnosis of an APG tumor, its location is a very important clue. The APG is composed of isolated normal salivary tissue, typically located approximately 6 mm anterior to the main parotid gland, between the skin and the masseter muscle, along an imaginary line that extends from the tragus to a point midway between the ala of the nose and the vermilion border of the lip [4, 6] (Figure 2(b)). The APG has a secondary duct emptying into Stensen’s duct. Although the APG is seldom mentioned in the literature, it exists in 21–69% of individuals [1,10]. The incidence of APG tumors reportedly ranges from 1% to 7.7% [2,6]. When our patient presented with a small lump in her right cheek, an APG tumor was suspected based on the location of the lesion. A retrospective review of her MRI study 3 years before her current presentation revealed that a small APG tumor was present at the time she underwent a resection of her MPG tumor.

There are many pathological entities that can involve the central cheek region, including metastatic tumors, Kimura’s disease, and vascular lesions such as hemangioma and vascular malformations [5]. Among these, APG tumors are the most common. Ultrasonography, CT, MRI, and sialography are useful for detecting the size, shape, and location of these central cheek tumors. Kronenberg et al. [3] recommended the use of CT to establish anatomic relationships. Today, the best imaging technique currently available for defining soft tissue lesions is MRI with gadolinium contrast. Both lesion size and location can be readily differentiated with combined coronal and axial views of the MPG and APG.

Sialography was once used routinely but was only marginal successful in establishing diagnoses. We do not rely on the sialography because it is a difficult test to perform, and the results are often not informative. Fine-needle aspiration (FNA) is helpful to establish a diagnosis of cystic lesions, but its role remains controversial for solid masses [2]. Although FNA of solid cheek masses can provide definitive diagnostic information, there is concern that FNA of solid lesions can lead to the spread of malignant cells to surrounding tissue. Once tissue has been obtained, the pathologic diagnosis of parotid tissue can be difficult, particularly in cases of borderline malignancy. It has been estimated that 26%–50% of APG tumors are malignant [2,6]; the malignancy rate reported for MPG tumors is 18.5% [2]. We did not perform FNA of our patient’s solid mass lesion involving the APG.

We determined that the best treatment approach for our patient would be to completely excise the tumor with adequate tumor margins to minimize the risk of recurrence. Resection could be completed with a skin incision above the tumor; this approach is easy, quick, and safe if the tumor mass is sufficiently large [4]. The direct skin approach is easy but leaves a facial scar; Rodino et al. [8] also emphasized the high risk of potential injury to branches of the facial nerve with direct approach. In contrast, the oral approach does not leave a facial scar but can be challenging in the event of unexpected and excessive bleeding given the limited exposure to localize the bleeding source. Use of the oral approach also requires careful monitoring of the
facial nerve to prevent nerve injury [9]. Polayes et al. [7] concluded that the oral approach was “ill-advised.” The oral approach is reserved for skilled surgeons, comfortable with this technique. We do not recommend the direct or the oral approach.

In our experience, the standard parotidectomy approach is safe, effective, and cosmetically appealing for the surgical management of APG tumor. Lin et al. [5] described other approaches for APG tumor treatment, e.g., a standard face lift incision or an extension of the incision superiorly into the scalp hairline and inferiorly into the cervical crease, to gain more access. Johnson et al. [2] described a similar incision that “angulated” anteriorly toward the mass, providing better exposure.

Perzik et al. [6] also advocated the use of a standard parotidectomy-type incision for APG tumors, without the use of the anterior extension used by Johnson et al. These approaches reduce the risk of injury to the facial nerve and Stensen’s duct when compared to the direct cheek incision. Using a parotidectomy approach, the zygomatic and buccal branches of the facial nerve can be identified with a nerve stimulator by microscopy without dissection of the facial nerve in the MPG.

APG tumors usually present as an asymptomatic central cheek mass. Diagnosis can be challenging as small masses arising in the central cheek region are easily overlooked but may be more sinister than larger masses present in the same area. Diagnostic work-up should include a careful physical examination with further characterization of the mass by CT and MRI. In consideration of the delicate anatomy and high rate of malignancy in APG tumors [6], we recommend a standard parotid incision and removal of APG tumor with sufficient tumor margins and limited facial-nerve dissection for the successful management of APG lesions [8].

4. Conclusion

APG tumors usually present as an asymptomatic cheek mass and can be difficult to identify, especially when the lesion is located along the course of the Stensen’s duct. APG tumors are thought to have a greater malignancy potential than MPG tumors. Although the coincidental presence of the MPG and APG tumors is quite rare, this case shows the importance of identifying the smaller and potentially more sinister lesion involving the APG when it is present while preparing for the MPG tumor excision.

References