Title: Cavernous Hemangioma of the Accessory Parotid Gland

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Abstract

We report a rare case of a hemangioma arising from the accessory parotid gland. The patient, a 45-year-old female, complained of a right mid-cheek mass. Magnetic resonance imaging (MRI) showed a well-defined mass located in the right buccal space, anterior to the masseter muscle and adjacent to Stensen's duct. The mass had high T2-weighted signal intensity and showed strong patchy enhancement with gadolinium. This mass was surrounded by a common capsule with the accessory parotid gland. These findings indicated a hemangioma originating from the accessory parotid gland. The mass was completely removed by an intraoral approach without postoperative facial palsy, skin deformity, and difficulty in secreting saliva. Histological examination of the tumor revealed multiple, thin-walled, and dilated blood vessels, confirming the diagnosis of a cavernous hemangioma. MRI was extremely useful in diagnosing the mass as a hemangioma before surgery, clarifying relationships between the mass and adjacent structures and determining the surgical approach to the mass.

Key words: cavernous hemangioma, accessory parotid gland, intraoral approach, magnetic resonance imaging

Introduction

Neoplastic tumors originating in the accessory parotid gland are rare[1,2,3]. These tumors usually present as mid-cheek swelling, and surgical approaches are controversial. We report the case of a cavernous hemangioma arising from the accessory parotid gland, which was diagnosed by magnetic resonance imaging (MRI) before surgery. The tumor was successfully removed by an intraoral approach without postoperative complications.

Materials and Methods

A 45-year-old female visited our hospital complaining of a mass in her right mid-cheek. She was known to have had the mass for at least twenty years; its size increased gradually during this time.

Physical examination revealed a firm, nontender, and mobile subcutaneous mass that was three centimeters in diameter (Fig. 1). Ultrasonography showed a comma-shaped mass with a well-defined margin separate from the parotid gland. The mass contained numerous small hypoechoic chambers, and vascular flow was identified on Doppler mode. No calcifications were present in the mass. On MRI (Fig. 2), a well-defined mass was located in the right buccal space, anterior to the masseter muscle and adjacent to Stensen's duct. The mass was surrounded by a common capsule with the accessory parotid gland. It had low T1- and high T2-weighted

signal intensities. On gadolinium-enhanced T1-weighted images, the high intensity area spread in a patchy manner over time. Signal voids were not identified. These findings suggested that the mass was a hemangioma originating in the accessory parotid gland. Thus, we did not perform a fine needle aspiration biopsy of the mass.

The operation was performed by an intraoral approach under general anesthesia with endotracheal intubation through the right nasal cavity. We used NIM-Response®2.0 (Medtronic Inc., Minneapolis, Minnesota) for nerve monitoring to ensure that the branches of the facial nerve were not injured. As a guide to prevent damage to the duct during surgery, a bougie was inserted into Stensen's duct from its opening in the oral cavity. An incision was made on the right buccal mucosa. After cutting an anterior part of the buccinator muscle a dark blue mass was identified in the buccal space. This mass was successfully removed through the buccal mucosal incision without rupture of its capsule (Fig. 3). Stensen's duct was preserved. Next, the borders of the incised buccinator muscle were sutured, after which the buccal mucosa was sutured. The operation lasted for sixty-seven minutes. The mass was dark red, comma-shaped, and 30 × 15 × 15 mm in size (Fig. 4). Histological examination of the mass revealed multiple, thin-walled, and dilated blood vessels, confirming the diagnosis of a cavernous hemangioma. The postoperative course was satisfactory and uncomplicated; facial palsy, skin deformity, and difficulty in secreting saliva were not observed. Three years later, there is no evidence of

recurrence.

Discussion

Hemangiomas originating in the accessory parotid gland are extremely rare[2]. To our knowledge, this is the second case of a hemangioma emerging from the accessory parotid gland in an adult and the first case to be treated by an intraoral approach.

MRI is useful in demonstrating the extent of buccal space lesions[4,5]. Hemangiomas in the buccal space, reported in the literature, had high T2-weighted signal intensity and were enhanced by gadolinium[4,5]. In the present case, MRI revealed a mass with high T2-weighted signal intensity and strong patchy enhancement with gadolinium, indicating the presence of a hemangioma. Furthermore, MRI was useful for determining the surgical approach to the mass, clarifying relationships between the mass and adjacent structures such as Stensen's duct and the masseter muscle. Fine needle aspiration biopsy is usually useful for the preoperative histological diagnosis of tumors in the head and neck regions. However, we consider it unnecessary when MRI strongly suggests that the tumor is a hemangioma.

Generally, accessory parotid glands are located on the masseter muscle with the zygomatic and buccal branches of the parotid plexus of the facial nerve within the lobes and overlying them as a net-like covering[6]. Branches of the transverse facial artery are usually buried in the lobes of

the accessory glands[6]. Therefore, it is essential to understand this anatomy of the accessory parotid gland when determining a surgical approach to the mass in this area.

Surgical approaches to tumors arising from the accessory parotid gland include a mid-cheek skin incision overlying the tumor[1], an intraoral approach[1,3], a parotidectomy-type incision without a parotidectomy[1], a standard parotidectomy[1], and a facelift approach[2]. In view of aesthetic results, the intraoral approach to the accessory parotid gland is better than the other approaches involving skin incisions although it is difficult to provide a good exposure for tumor resection. We believe that a benign vascular lesion, as that in the present study, can be a good candidate for an intraoral approach because it allows resection extremely close to its capsule and minimizes damage to adjacent structures such as the facial nerve, Stensen's duct, and blood vessels. However, we do not recommend this approach when the tumor is preoperatively suspected to be a pleomorphic adenoma or malignant tumor because resection of such tumors generally requires a good exposure and sufficient surgical margins, and their capsules should not be ruptured during resection.

We believe that the intraoral approach can be useful when the tumor meets the following conditions: 1. preoperative diagnosis as a benign vascular lesion such as a hemangioma, 2. not an extremely large size (<3 cm), and 3. located on the anterior portion of the masseter muscle (not too far from the buccal mucosa). Furthermore, it is ideal if nerve monitoring is performed

to avoid facial nerve injuries.

References

1. Johnson FE, Spiro RH. Tumors arising in accessory parotid tissue. *Am J Surg* 1979;138:576-578.

2. Chang CH, Mun GH, Lim SY, et al. Cavernous vascular tumor of the accessory parotid gland. *J Craniofac Surg* 2007;18:1493-1496.

3. Schmutzhard J, Schwentner IM, Andrle J, et al. Resection of accessory parotid gland tumors through a peroral approach with facial nerve monitoring. *J Craniofac Surg* 2007;18:1419-1421.

4. Tart RP, Kotzur IM, Mancuso AA, et al. CT and MR imaging of the buccal space and buccal space masses. *Radiographics* 1995;15:531-550.

5. Kurabayashi T, Ida M, Tetsumura A, et al. MR imaging of benign and malignant lesions in the buccal space. *Dentomaxillofac Radiol* 2002;31:344-349.

6. Toh H, Kodama J, Fukuda J, et al. Incidence and histology of human accessory parotid glands. *Anat Rec* 1993;236:586-590.

Figure legends

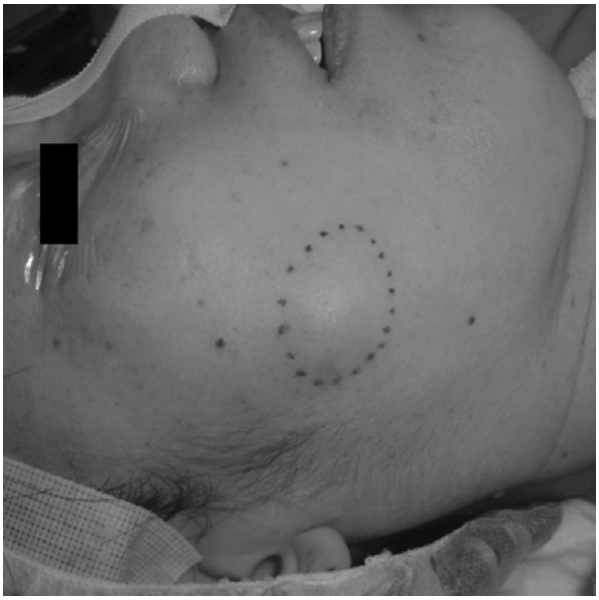

Fig. 1 Preoperative view. The dotted circle indicates tumor margins.

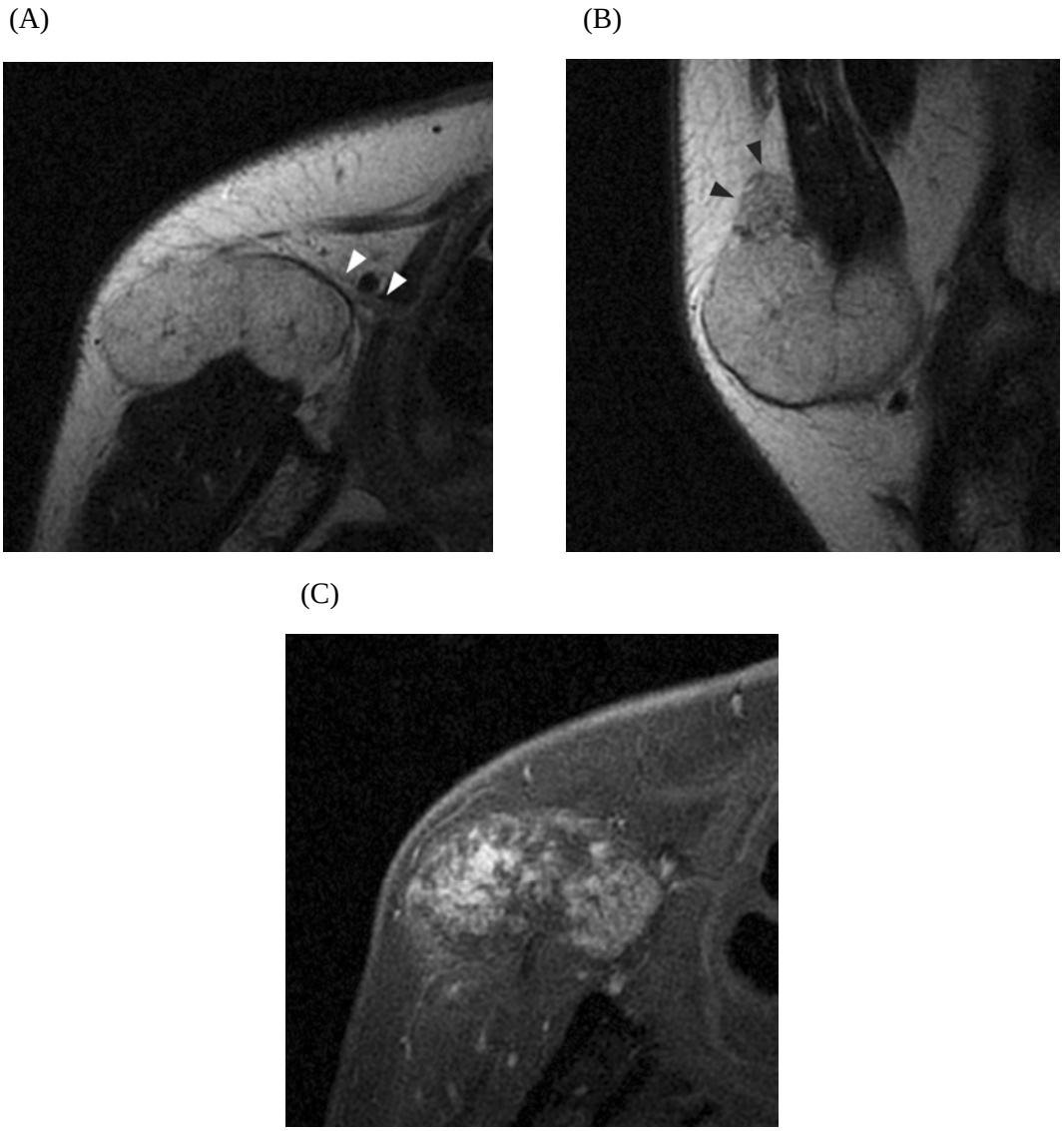

Fig. 2 MRI. (A) T2-weighted axial image. A well-defined mass with high signal intensity was located in the right buccal space anterior to the masseter muscle. The mass and Stensen's duct (white arrows) were in contact with each other. (B) T2-weighted coronal image. The mass was surrounded by a common capsule with the accessory parotid gland (black arrows). (C) Gadolinium diethylenetriamine-pentaacetic acid enhanced the T1-weighted image with fat suppression. The mass showed a strong patchy enhancement.

Fig. 3 Intraoperative view. The mass appeared from the incision in the right buccal mucosa.

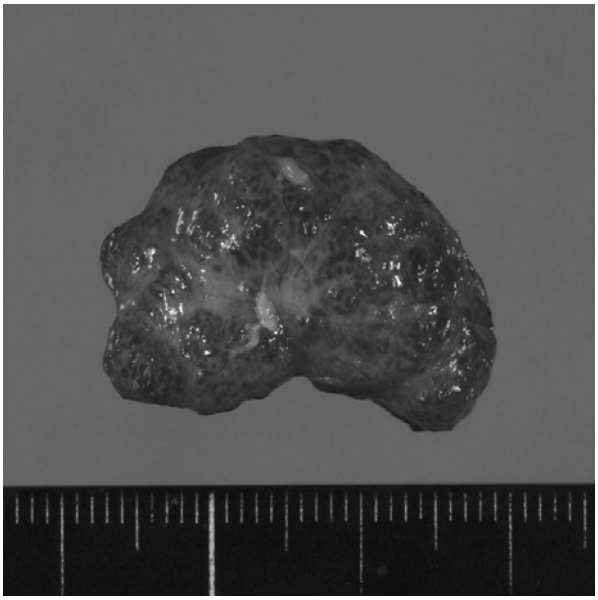

Fig. 4 Macroscopic findings of the tumor.