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CASE REPORT

Temporal Bone Invasion by Recurrent Benign Pleomorphic Adenoma of the Parotid Gland

Yasuomi Kunimoto, Filippo Di Lella, Shailendra Sivalingam, Giuseppe De Donato, Maurizio Falcioni, Mario Sanna

Department of Otolaryngology and Head and Neck Surgery, Tottori University, Yonago, Japan (YK)
Gruppo Otologico, Piacenza-Roma, Italy (YK, FDL, SS, GDD, MF, MS)

Objective: We present a case report of a 55-year-old woman who had multiple recurrences with upward extension along the fallopian canal via the stylomastoid foramen.

Results: Pleomorphic adenoma is the most common neoplasm of the parotid gland, and is associated with the risk of recurrence. The spread of parotid tumors into the temporal bone is uncommon. The tumor was found to be centered around the stylomastoid foramen, extending to the hypotympanum, infralabyrinthine cells, and close to the inferior wall of the internal auditory canal. Complete surgical resection of the recurrent tumor was performed by a subtotal petrosectomy with upper neck extension.

Conclusion: The surgical approach should provide excellent visibility with a wide surgical exposure to secure local neurovascular structures. The existence of recurrent or metastasizing pleomorphic adenoma underscores the need for meticulous and complete surgical excision of the primary tumor, and close clinical follow-up.

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Introduction

Pleomorphic adenoma is the most common neoplasm of the parotid gland. It is associated with the risk of recurrence and malignant transformation. Tumor spillage, incomplete excision, and violation of the tumor pseudocapsule are considered the only proven factors contributing to recurrent disease[1]. The upward spread of recurrent pleomorphic adenoma along the fallopian canal via the stylomastoid foramen is uncommon.

We present one such case of regional spread of recurrent pleomorphic adenoma to the temporal bone.

Case report

A 55-year-old woman was referred to our department in 2011. She had a history of pleomorphic adenoma of the left parotid gland, for which surgery was first performed in 1983. Furthermore, she had experienced three recurrences and excisions elsewhere: in 2000, 2001, and 2009. The details of the surgeries performed in 1983, 2000, and 2001 were unclear, but total parotidectomy was performed in 2009. Additionally, she had developed a subcutaneous mass in the gluteal region, which was resected in 2009. In all these instances, including the gluteal mass, pathologic analysis showed pleomorphic adenoma without malignancy. Physical examination showed a left facial paralysis rated as House-Brackmann grade VI; this had been present since the surgery in 2001. The remainder of the clinical examination, including otological inspection and audiometry, was unremarkable. Computed tomography (CT) revealed a solid mass, measuring 18mm x 26mm in the left mastoid (Fig. 1), with no evidence of cervical lymphadenopathy.

Corresponding address:
Yasuomi Kunimoto
Department of Otolaryngology and Head and Neck Surgery, Tottori University
86 Nishi-cho, Yonago-shi, Tottori, JAPAN
Tel: +81-859-38-6627
Fax: +81-859-38-6629
E-mail address: kunimon0608@yahoo.co.jp

On magnetic resonance imaging (MRI), the lesion was hyperintense on T2-weighted images and isointense on T1-weighted images with enhancement after gadolinium infusion. Total surgical resection was performed with a subtotal petrosectomy and upper neck extension. The skin incision was extended to the upper neck and the internal jugular vein was identified. In the event of bleeding from the jugular bulb during the petrosectomy, the internal jugular vein could be ligated and the sigmoid sinus occluded with oxidized cellulose (Surgicel, Ethicon). During surgery, the tumor was in contact with the superior and lateral wall of the jugular bulb, and had eroded the posterior wall of the external auditory canal. Additionally, the tumor was found to be centered around the stylomastoid foramen, extending to the hypotympanum, infralabyrinthine cells, and close to the inferior wall of the internal auditory canal. The tumor comprised a single multiloculated, unencapsulated mass. It circumferentially involved the facial nerve from the 2nd genu to the stylomastoid foramen. In view of the preoperative grade VI facial paralysis since 2001, we proceeded with resection of the facial nerve without any reconstruction. As a result, a meticulous total resection was achieved with carefulness that allowed the preservation of the jugular bulb. The defect was obliterated using abdominal fat. Pathologic analysis of the resected specimen showed pleomorphic adenoma without evidence of malignancy or carcinoma ex pleomorphic adenoma. No sign of recurrence has been found during 12 months' follow-up (Fig. 2).

Discussion

Pleomorphic adenoma is the most frequent parotid gland tumor, and is associated with a high rate of recurrence despite being characterized as a benign neoplasm. As for our case, because the details of the surgeries performed previously were unclear, there was a possibility that the tumor contained an in-situ carcinoma or had minimal invasion. In view of the recurrence after total parotidectomy performed at another institution, a complete resection in the first surgery is the most important for pleomorphic adenoma.

Perineural or extraneural spread of benign parotid tumors into the temporal bone is not a well-documented occurrence. The extension of tumor into the stylomastoid foramen may be more likely to occur in patients who have had previous parotid surgery. Possible explanations include fibrous scar tissue formation in the parotid wound bed which could block spread into the neck or face and favor extension into the nonviolated stylomastoid foramen and temporal bone[2]. These tumors are less likely to be detected early because the tumor enlarges in the mastoid cavity without any symptoms. Although it is common for malignant tumors to spread in this way, this type of spread is rare for benign parotid tumors such as pleomorphic adenoma. Accordingly, facial nerve dysfunction, although extremely rare, can be the result of benign parotid tumors.

Apart from spreading in this manner, Peters et al[3]. reported pleomorphic adenoma of the middle ear and mastoid, existing in isolation from the original salivary glands. They concluded that the ectopic salivary gland tissue within the middle ear cleft was the most probable origin of their tumor. This is very unlikely in our case and we are of the opinion that here it is more likely due to perineural or extraneural spread.

The surgical treatment of recurrent pleomorphic adenoma has never been standardized. The type of surgery must be individualized to consider the extent of previous surgery, the type of recurrence, and the patient's age[4]. With surgical excision, prognosis in these patients are generally favorable. In our case, we had prepared the upper neck extension to provide optimal control of the jugular foramen region, because the present recurrence had exposed a large surface of the jugular bulb. If significant bleeding occurs from the jugular bulb, closure of the sigmoid sinus and internal jugular vein is required[5]. A wide resection including all possible pathways of tumor spread from the stylomastoid foramen is required. According to the size or position of the tumor, a translabyrinthine approach or infratemporal fossa approach type A should be prepared. However, we were able to resect the entire tumor and surrounding tissue via a subtotal petrosectomy facilitating preservation of the jugular bulb.

Some authors suggest that radiotherapy should be reserved only for malignant tumors. Nonetheless, its use is not infrequent[6,7]. Jackson et al.[6] used postoperative radiotherapy in patients in whom histology showed that resection margins of the specimen were not free of tumor or in those with intraoperative spillage of tumor. Renehan et al.[7] recommended postoperative radiotherapy in multinodular recurrences. Although Redaelli de Zinis et al[8]. showed no significant

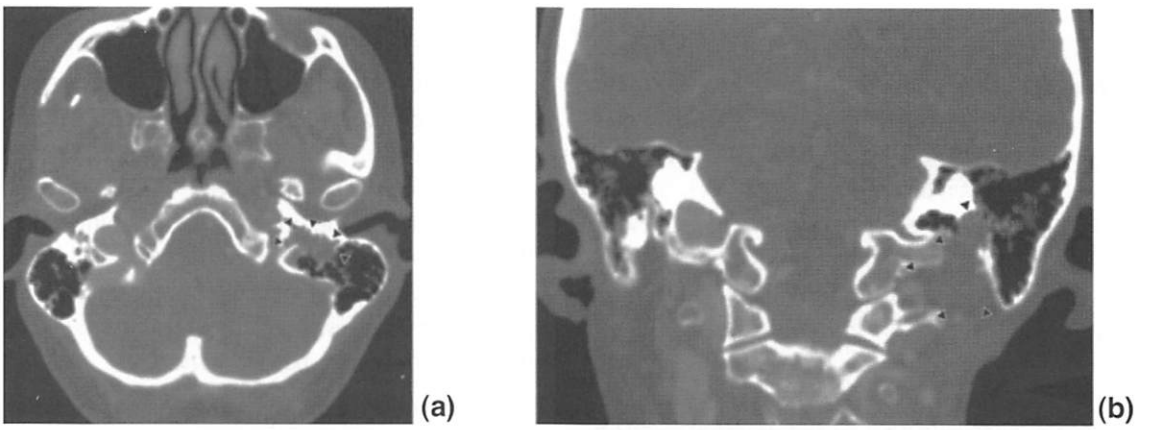

Figure 1. Computed tomography (CT) shows tumor extending into the temporal bone along the route of the facial nerve from stylomastoid foramen (arrow heads, a; axial, b; coronal).

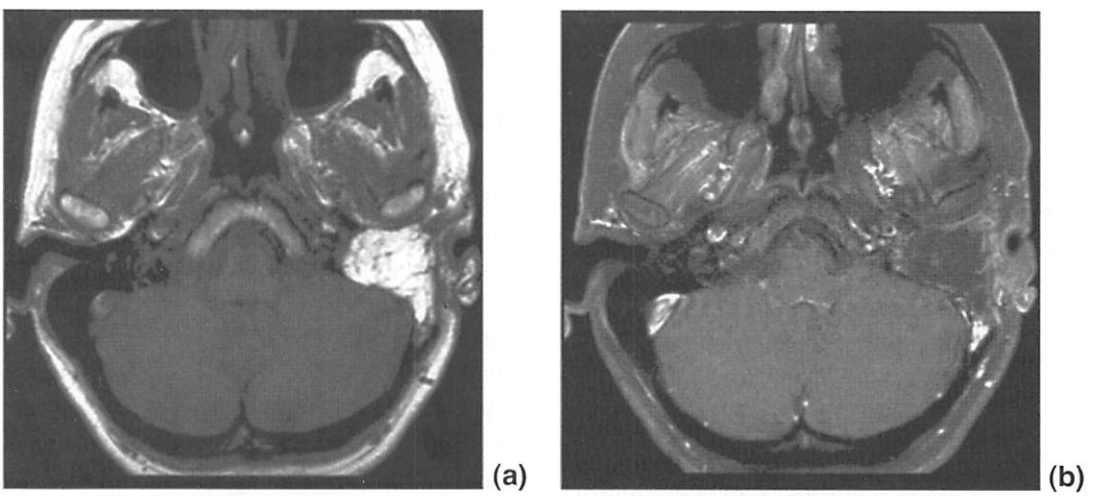

Figure 2. Postoperative axial MRI shows no sign of recurrence. The defect is obliterated using abdominal fat. Fat suppression technique is useful to reveal the adjacent pathology. (a; T1-weighted, b; T1-weighted with fat suppression).

association between postoperative radiation and recurrence rate, their cases were too limited to draw any conclusions about indications for radiation therapy. Although our case had experienced multiple recurrences, postoperative radiation was not considered this time because the tumor could be resected completely with the surrounding tissue.

Pleomorphic adenoma is an epithelial tumor that originates from glandular tissue and can be seen not only in salivary glands, but also in skin appendages or other glandular tissue. Although recurrence can be recognized as a feature of benign tumors, and metastasis as a feature of malignant tumors, since the 1940s, a relatively unique situation has been noted in which a benign pleomorphic adenoma without malignant transformation inexplicably metastasizes to distant sites[9]. This has given rise to the term 'metastasizing pleomorphic adenoma' which although a pathological contradiction, describes a situation where metastasis occurs from a benign pleomorphic adenoma in the absence of a malignant tumor. Ikeda et al[10]. resected a bone tumor 44 years after resection of a primary submandibular gland tumor and indicated the possibility of metastasis using genetic analysis. The mechanism underlying this metastatic behavior remains unclear, but hypotheses include vascular or lymphatic invasion by the primary tumor, or hematogenous spread from the surgical manipulation[11]. As for our case, in the absence of detailed pathological

reports and a genetic comparison, we cannot be certain that the gluteal tumor resected at a previous institution 26 years after the first primary tumor resection is a metastasis. Nevertheless, 12 months' follow-up is insufficient and long-term monitoring is mandatory after treatment.

Conclusion

Clinicians should be aware that benign pleomorphic adenomas, are associated with the risk of recurrence and also metastasis. MRI can localize the tumor and can guide the surgeon in planning the right approach. The surgical approach should provide excellent visibility with a wide surgical exposure to secure local neurovascular structures. Facial nerve resection can be considered in cases of multiple recurrences and preoperative facial weakness. The existence of recurrent or metastasizing pleomorphic adenoma underscores the need for meticulous and complete surgical excision of the primary tumor, and close clinical follow-up.

Summary

The upward spread of recurrent pleomorphic adenoma along the fallopian canal via the stylomastoid foramen is uncommon.

Additionally, metastasizing pleomorphic adenoma is also uncommon.

We present one such case of regional spread of recurrent pleomorphic adenoma to the temporal bone with the possibility of gluteal metastasis.

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Erratum

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The last author's name Melin Cayonu have to be corrected as Melih Cayonu