

Sebaceous Carcinoma of the Parotid Gland: A Case Report

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Key Words

Sebaceous carcinoma · Parotid gland · Surgery

Abstract

Background: Primary sebaceous carcinoma of the parotid gland is extremely rare, and because of its rarity, clinicopathological characteristics and histogenesis are not fully understood. **Methods:** Here, we report a patient who presented with a left infra-auricular painless mass. We present the histological features and discuss possible optimal treatments based on previous literature. **Results:** The mass was suspected to be a myoepithelial tumor or possibly a pleomorphic adenoma. Initially, the mass was resected with preservation of the facial nerve, but this caused facial palsy. Because the histological examination showed a sebaceous carcinoma and a part of the mass could be remaining on the facial nerve, additional surgery was performed, and the facial nerve was reconstructed with cervical nerve. Follow-up after 7 months showed no sign of recurrence of metastasis. **Conclusion:** We encountered a rare sebaceous carcinoma of the parotid gland. Additional surgery was performed because pre-operative diagnosis was difficult.

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Background

The sebaceous gland can sometimes be detected in normal major salivary glands, but sebaceous cell-containing salivary gland neoplasms are rare. Considering the salivary glands, most cases involve the parotid gland and are rarely reported in the other major glands or intraorally [1–4]. In the English literature, only 24 cases of sebaceous carcinoma (SC) in the parotid gland have been reported [5–17]. SC in the parotid gland has a bimodal age distribution, with a peak in the second decade and another one in the seventh decade of life (with a range of 6–92 years). This is also the age distribution of sebaceous cells which appear in the

normal salivary gland and the adjacent regions. They are rarely seen in children under the age of 10 years [6] but appear to increase in numbers in the second decade of life. It has been suggested that this may reflect the same hormonal stimulus that activates the sebaceous glands in the skin during puberty. SC etiology is unknown, but the male-to-female ratio is 3:5. The patients often present with a painless, slow-growing, asymptomatic swelling, but some experience pain, and a few cases with facial paralyses have been reported. Here, we report a case of SC in the parotid gland with a brief review of the literature and discussion of possible appropriate treatments.

Case Presentation

In January 2008, a 75-year-old male presented with a left infra-auricular painless mass, which he had had for 1 year. The mass was suspected to be a myoepithelial tumor after fine needle aspiration (FNA) biopsy at another hospital. He was then sent to our hospital for further assessment. Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Upon physical examination, the size of the mass was about 3 cm, and it was located at the superior side of the left parotid gland. The patient had no complaints of pain, tenderness, or facial palsy. The mass was elastic and firm, and mobility was good. Echograms showed that the size of the mass was 33 × 30 × 24 mm, with a relatively regular border. Back echoes were enhanced and internal echoes were dissimilar. The blood flow of the mass was examined by Doppler echograms, and the results showed that it was poor (fig. 1). Magnetic resonance imaging (MRI) showed a hypo-isointense lobular mass in the superior part of the left parotid gland on T1-weighted images (fig. 2a, d). The mass was slightly enhanced (fig. 2c), and T2-weighted images showed high intensity in the region (fig. 2b). The mass presented within the parotid gland, and lymphadenopathy was not detected in the examined region. FNA was performed twice but failed to detect any valuable cells. Even though we could not exclude the possibility of a low-grade malignancy tumor, pleomorphic adenoma was highly suspected from the MRI findings and the clinical features. We proposed a surgical treatment to the patient but he denied it.

He had pain in the region of the mass 2 months after the first examination, which made him decide to undergo the surgical treatment. At the time, he rejected our proposal of re-examination by FNA. In May 2009, we resected a tumor of the left parotid gland. A tumor existed within the superficial parotid gland without a capsule and adhered to the main branch of the facial nerve but not to the surrounding tissues, such as the subcutaneous connective tissues and the sternocleidomastoid muscle. The mass was carefully detached from the facial nerve and then resected with the superficial parotid gland. The mass appeared of a yellow-white color and formed lobular nodules involving the parotid gland (fig. 3a). Intraoperative rapid frozen pathology diagnosed it as a malignant tumor but could not identify the exact tissue type. Even with careful handling, left facial movement was almost completely paralyzed immediately after the surgery (House-Brackmann score: grade V).

Pathologic findings showed that the tumor cells had two cell populations. One had small rounded nuclei with eosinophilic cytoplasm and the other had vacuolar cytoplasm with peripherally located nuclei (fig. 3b). These cells mixed and formed nests which had prominent nucleoli and underwent mitosis. These tumor cells were unable to be stained with periodic acid-Schiff (PAS) stain (fig. 3c) and PAS with diastase (D-PAS) (fig. 3d) but strongly stained with Oil Red O (fig. 3e), which identified the tumor as a SC.

Because the tumor could have possibly remained at the site of the facial nerve where it was detached, additional surgery was proposed, and the patient agreed to have it. In June 2009, 1 month after the first operation, we performed total left parotidectomy with removal of a part of the main branch of the facial nerve. The facial nerve was reconstructed by a cervical nerve connecting the main branch to each of the peripheral branches. Left selective neck dissection was also performed. Upon histological assessment no tumor was detected in the additional removed tissues. Because SC can be a part of the Muir-Torre syndrome, gastrointestinal endoscope and colon fiberscope were examined, and no tumor was found in the duct. Postoperative radiation therapy was not selected, and the patient is doing well with no metastasis or recurrences at 7 months after the second surgery.

Discussion

A sebaceous cell is a vacuolated clear cell involved mainly with the eyelids and occasionally with other skin sites and the normal parotid glands, submandibular glands, and sublingual glands [18]. Sebaceous adenoma, sebaceous lymphadenoma, SC, and sebaceous lymphadenocarcinoma originated from sebaceous cells are rarely tumorous. Though the origin of SC within the major salivary glands is unclear, SC has mainly been reported in the parotid glands. Because of its rarity, clinicopathological characteristics and histogenesis are not fully understood. The sebaceous cell is found to peak in the second and seventh decades of life, which parallels to the age of SC patients reported. SC is a rare malignant tumor, and only 24 cases of SC in the parotid gland have been reported in the English literature [5–17]. The present case brings the total number of recorded cases to 25.

Generally, SC in the parotid gland presents as painless and slow growing, which results in tender swelling for several months or even years. Facial nerve palsy and pain in the mass is usually a late event, and the mass is occasionally fixed to the skin. Metastasis may occur in the lung, brain, and regional lymph nodes.

Usually, a parotid tumor is evaluated by various examinations such as echograms, CT, MRI, FNA biopsy, radionuclide salivary scan imaging, and positron emission tomography. Because several kinds of malignant tumors arise in the parotid gland, imaging examinations alone cannot lead to the histological diagnosis. It is known that FNA is suitable to diagnose a benign tumor, but it is difficult to evaluate the exact histology in malignant tumors [19]. In this case, a myoepithelial tumor was suspected by the preoperative FNA, but we considered a pleomorphic adenoma based on the MRI findings and the clinical features. When the patient was referred to our hospital with pain in the left parotid region, a tumor was suspected as malignant change of pleomorphic adenoma or other malignant tumors. We proposed to perform FNA again, but he refused.

The SC slightly simulates mucoepidermoid tumors. Several reports have confirmed the usefulness of histochemistry as well as immunohistochemistry to diagnose as SC [6, 20]. However, PAS and D-PAS stains were negative, confirming that the vacuolated clear cells had no glycogen, which indicated that the cells were not mucoepidermoid cells. Strong stains of Oil Red O revealed that the tumor cells had a feature of SC. The tumors are composed predominantly of sebaceous cells of varying maturity arranged in nests and/or sheets with different degrees of atypia, pleomorphism, and invasiveness.

SC in the parotid gland is classified as a moderate malignant carcinoma according to the 2005 WHO classification of salivary gland tumors. The rarity of the neoplasm means that the optimal treatments have yet to be established. In the reported cases, surgical treatment was performed in most cases. Seven out of the 19 operative cases reported (36.8%) have result-

ed in local recurrence and/or lymph node metastasis [5, 7, 11, 13, 16]. Out of the 19 operative cases, 9 were treated with radiotherapy considered as an adjuvant therapy [5–7, 9, 11, 13] and only 1 case was performed as preoperative irradiation [14]. Though the tumor progression was unclear in the reported cases, only 1 case treated with radiotherapy recurred. This indicates that radiotherapy was a valuable treatment as adjuvant therapy in parotid SC. Four cases were treated with chemotherapy [5]; however, 2 cases (70%) progressed to metastasis. Because 5 out of 7 recurrent cases died within an average of 3.8 years, appropriate treatment was critical at the primary stage. Of the cases reported, 19 were treated by local excision and 7 resulted in recurrence. Since the mortality rate of the recurrence cases was nearly 50%, it is better to choose parotidectomy, extended parotidectomy, and/or neck dissection to achieve complete resection. However, in the case of recurrence after local excision, SC was successfully removed by salvage operation [11], which shows how difficult it is to establish the standard treatment.

In the present case, we performed additional surgery because the tumor strongly adhered to the facial nerve by visual assessment, and we could not confirm a complete resection by completion of histology after the first operation. As a result, we did not confirm the presence of the tumor in additional tissues histologically. Although SC in the parotid gland is a moderate-grade malignant tumor, it recurs easily and the mortality rate is high when recurrence occurs, so we believe radical excision was appropriate when we diagnosed the tumor as SC.

Conclusion

We experienced a rare SC of the parotid gland. Additional surgery was performed because preoperative diagnosis was difficult. Histologic features were presented and possible optimal treatment was discussed based on previous literature.

Disclosure Statement

The authors declare that they have no competing interests.

References

- 1 Alawi F, Siddiqui A: Sebaceous carcinoma of the oral mucosa: case report and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2005;99:79–84.
- 2 Esnal Leal F, Garcia-Rostan y Perez GM, Garatea Crelgo J, Gorriaran Terreros M, Arzoz Sainz de Murieta E: Sebaceous carcinoma of salivary gland. Report of two cases of infrequent location (in Spanish). *An Otorrinolaringol Ibero Am* 1997;24:401–413.
- 3 Ohara N, Taguchi K, Yamamoto M, Nagano T, Akagi T: Sebaceous carcinoma of the submandibular gland with high-grade malignancy: report of a case. *Pathol Int* 1998;48:287–291.
- 4 Diedhiou A, Cazals-Hatem D, Rondini E, Sterkers O, Degott C, Wassef M: Sebaceous carcinoma of the submandibular gland: a case report (in French). *Ann Pathol* 2001;21:348–351.
- 5 Akhtar M, Gosalbez TG, Brody H: Primary sebaceous carcinoma of the parotid gland. *Arch Pathol* 1973;96:161–163.
- 6 Altemani A, Vargas PA, Cardinali I, Aguiar SS, Lopes M, Soares AB, et al: Sebaceous carcinoma of the parotid gland in children: an immunohistochemical and ploidy study. *Int J Oral Maxillofac Surg* 2008;37:433–440.
- 7 Gnepp DR, Brannon R: Sebaceous neoplasms of salivary gland origin. Report of 21 cases. *Cancer* 1984;53:2155–2170.
- 8 Granstrom G, Aldenborg F, Jeppsson PH: Sebaceous carcinoma of the parotid gland: report of a case and review of the literature. *J Oral Maxillofac Surg* 1987;45:731–733.

- 9 Grieve TP, Saragas E, Mannell A: Sebaceous carcinoma of the parotid gland in a black patient. A case report. *S Afr Med J* 1986;69:141–142.
- 10 Linhartova A: Sebaceous cells in repeat biopsies of normal salivary glands and in sebaceous carcinoma of the parotid gland (in Czech). *Cesk Patol* 1982;18:21–25.
- 11 MacFarlane JK, Vilorio JB, Palmer JD: Sebaceous cell carcinoma of the parotid gland. *Am J Surg* 1975;130:499–501.
- 12 Mighell AJ, Stassen LF, Soames JV: Sebaceous carcinoma of the parotid gland. *Dentomaxillofac Radiol* 1996;25:51–53.
- 13 Shulman J, Waisman J, Morledge D: Sebaceous carcinoma of the parotid gland. *Arch Otolaryngol* 1973;98:417–421.
- 14 Silver H, Goldstein MA: Sebaceous cell carcinoma of the parotid region. A review of the literature and a case report. *Cancer* 1966;19:1773–1779.
- 15 Siriwardena BS, Tilakaratne WM, Rajapakshe RM: A case of sebaceous carcinoma of the parotid gland. *J Oral Pathol Med* 2003;32:121–123.
- 16 Takata T, Ogawa I, Nikai H: Sebaceous carcinoma of the parotid gland. An immunohistochemical and ultrastructural study. *Virchows Arch A Pathol Anat Histopathol* 1989;414:459–464.
- 17 Tsukada Y, Delapava S, Pickren JW: Sebaceous-cell carcinoma arising in mixed tumor of parotid salivary gland. report of a case. *Oral Surg Oral Med Oral Pathol* 1964;18:517–522.
- 18 Batsakis JG, Littler ER, Leahy MS: Sebaceous cell lesions of the head and neck. *Arch Otolaryngol* 1972;95:151–157.
- 19 Zbaren P, Schar C, Hotz MA, Loosli H: Value of fine-needle aspiration cytology of parotid gland masses. *Laryngoscope* 2001;111:1989–1992.
- 20 Hasebe T, Mukai K, Yamaguchi N, Ishihara K, Kaneko A, Takasaki Y, et al: Prognostic value of immunohistochemical staining for proliferating cell nuclear antigen, p53, and c-erbB-2 in sebaceous gland carcinoma and sweat gland carcinoma: comparison with histopathological parameter. *Mod Pathol* 1994;7:37–43.

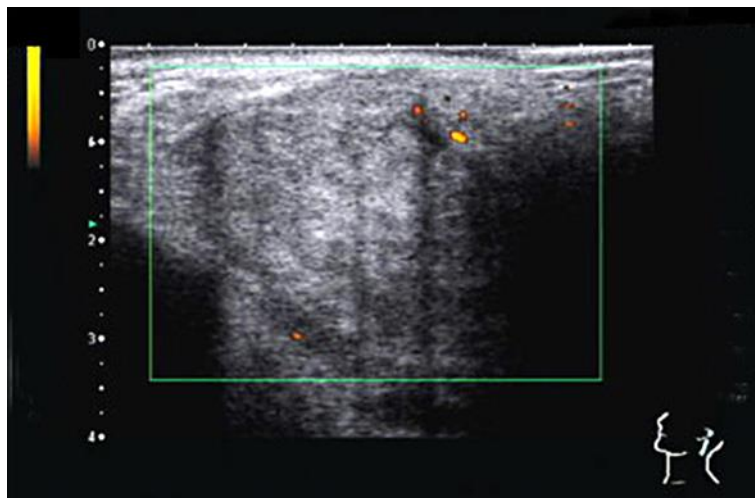


Fig. 1. Doppler echogram of the left parotid gland. The mass showed a relatively regular border, enhanced back echoes, dissimilar internal echoes, and poor blood flow.

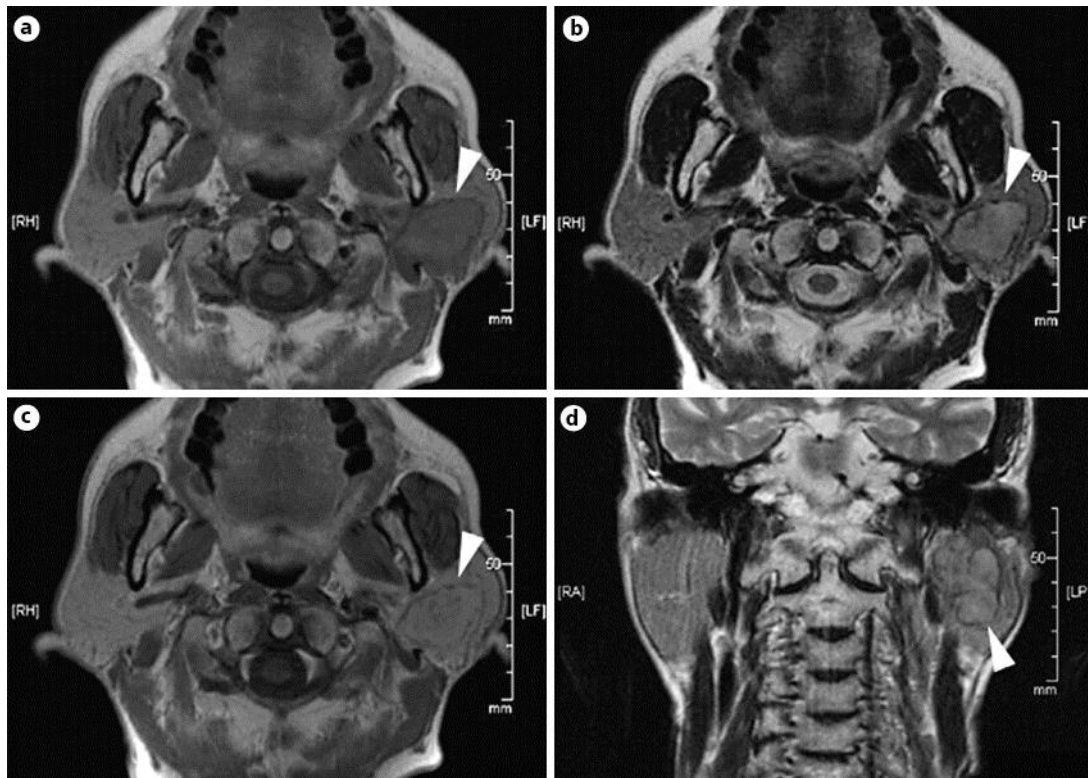


Fig. 2. MRI showed a lobular hypo-intense mass on T1-weighted images (arrowhead in **a**), high intensity on T2-weighted images (arrowhead in **b**), and was well enhanced (arrowhead in **c**) in the superior part of left parotid gland (arrowhead in **d**).

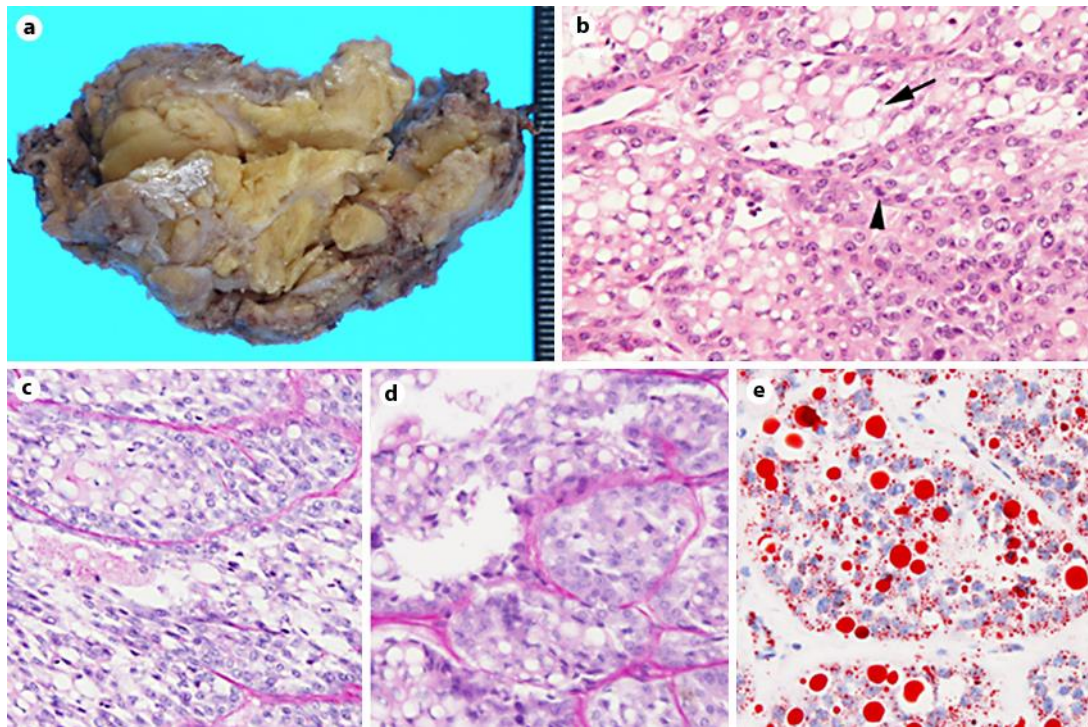


Fig. 3. **a** SC of the left parotid gland showing lobular nodules in the gland. **b** SC showing two cell populations of small basophilic cells (arrowhead) and vacuolated cytoplasm (arrow). HE. Original magnification. $\times 100$. **c** SC cells were negative for PAS stains. HE. Original magnification. $\times 400$. **d** SC cells were negative for D-PAS stains. HE. Original magnification. $\times 400$. **e** SC cells were positive for lipids with Oil Red O (HE. Original magnification. $\times 400$).