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CASE REPORT

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MUCOEPIDERMOID CARCINOMA OF STENSEN'S DUCT: A CASE REPORT AND REVIEW OF THE LITERATURE

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Abstract: *Background.* Mucoepidermoid carcinoma of Stensen's duct is a rare neoplasm, with only five cases reported in the literature.

Methods. We report another case of mucoepidermoid carcinoma of Stensen's duct and review the literature.

Results. Stensen's duct neoplasms tend to be symptomatic at an early stage by causing an obstruction of the parotid duct. New imaging techniques such as MR sialography and sialoendoscopy are very helpful in diagnosis and patient management.

Conclusions. Although the rarity of this condition prevents definitive conclusions about the optimal treatment, we propose that Stensen's duct neoplasms should be treated like similar neoplasms occurring in the parotid gland tissue, taking into consideration clinical stage, tumor grade, and surgical margins.
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Keywords: mucoepidermoid carcinoma; Stensen's duct; parotid gland; sialoendoscopy; MR sialography

Primary malignant tumors of Stensen's duct are very rare, with a total of 24 cases reported in the literature. Because of the anatomic course of Stensen's duct, tumors occurring in this duct can be easily confused with those of the minor salivary glands of the oral cavity, the accessory parotid gland, or the parotid gland itself.[1] Mucoepidermoid carcinoma is the most common malignancy of the salivary glands, occurring more frequently in the major salivary glands and most frequently in the parotid gland. Only five cases of mucoepidermoid carcinoma occurring in Stensen's duct have been reported in the literature.[2–4]

In this article, we present a case of a primary mucoepidermoid carcinoma of Stensen's duct in a 61-year-old woman, and we systematically review the literature, hoping to shed some light on the management and prognosis of this rare tumor entity.

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CASE REPORT

We present a case of a 61-year-old woman, who consulted our clinic for an intermittently painful mass in the left cheek for the previous year. Her medical history was not significant. Physical examination revealed a firm, subcutaneous, 1.5-cm mass located below the left zygomatic arch, fixed to the underlying tissue but not to the skin.

There was no palpable cervical lymphadenopathy, no facial weakness, and no noticeable abnormalities within the oral cavity. Sonography and MRI revealed the presence of a mass measuring 1.5 × 0.5 × 0.5 cm between the skin and the masseter muscle, with no signs of soft tissue infiltration (Figure 1). MR sialography showed an absence of signal in Stensen's duct for about 2 cm associated with signs of dilation of the intraglandular parotid duct system (Figure 2). The suspicion of intraductal disease led us to complete the investigation by performing a diagnostic sialoendoscopy, which revealed a mass situated approximately 3.5 cm proximal to the papilla and fixed to the ductal wall, with complete obstruction of Stensen's duct, findings very suspicious of a tumor of the parotid duct.

A surgical procedure was performed, including a superficial parotidectomy, an excision of the entire Stensen's duct, and the sacrifice of two small distal midfacial nerve branches and portions of the adjacent masseter muscle. Intraoperative frozen section yielded a diagnosis of low-grade mucoepidermoid carcinoma. The deep parotid lobe was left intact, and no neck dissection was undertaken.

Sections from the surgical specimen were examined with routine hematoxylin-eosin staining. On microscopic examination, we observed a tumor measuring 1.5 × 0.5 × 0.5 cm arising from Stensen's duct wall and protruding in the lumen, without extension beyond the duct wall (Figure 3). This tumor consisted of two cell types, mucus-secreting cells and squamous cells (Figure 4). Mayer's mucicarmine stain highlighted the mucus within the cells. A discrete cellular atypia, rare mitotic figures, and absence of necrosis were noted. The histologic diagnosis was consistent with low-grade mucoepidermoid carcinoma arising in the Stensen's duct and confirmed clear surgical margins. Thus, the clinical and pathologic classification was T1N0M0.[5] The patient did not undergo further therapy. Postoperative controls showed a slight temporary partial facial

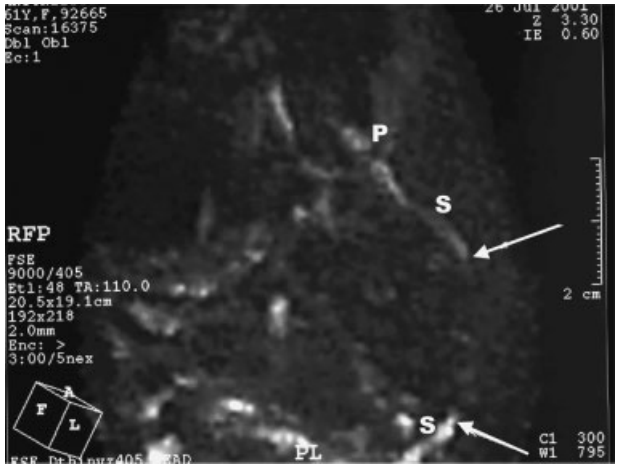

FIGURE 2. MR sialography showing main parotid duct (S) with complete obstruction (arrows) located approximately 3.5 cm proximal to the papilla (P).

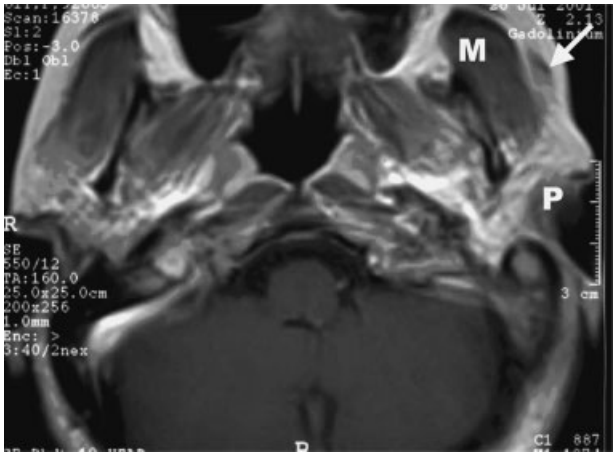

FIGURE 1. Gadolinium-enhanced, T1-weighted axial MR image through the parotid region, showing a 1.5- × 0.5- × 0.5-cm mass (arrow) situated between the skin of the left cheek and the masseter muscle. P, parotid gland; M, masseter muscle.

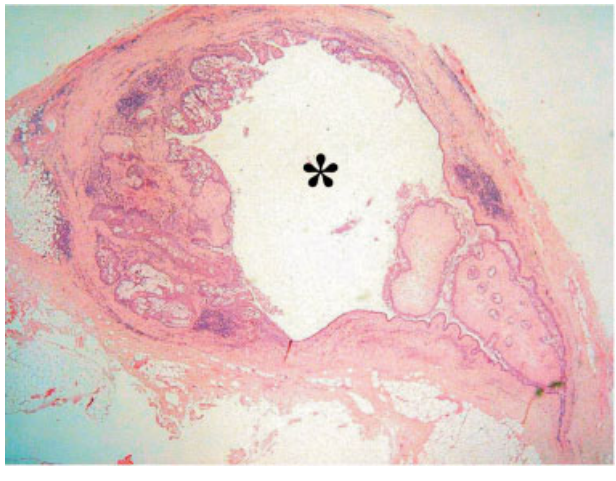

FIGURE 3. Microscopic examination of a cross section of Stensen's duct 4 cm proximal to the buccal mucosa shows a tumoral lesion arising from the ductal wall and protruding in the ductal lumen (star). Note that the tumor did not extend beyond the confines of the ductal wall (hematoxylin-eosin stain, original magnification ×10). [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

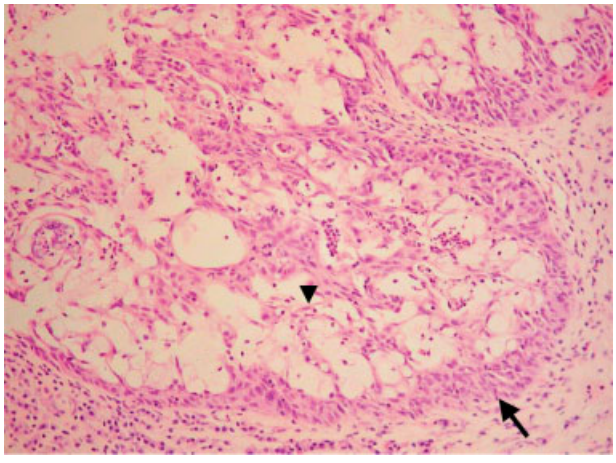

FIGURE 4. Low-grade mucoepidermoid carcinoma consisting of two cell types, mucus-secreting (arrowhead) and squamous cells (arrow) (hematoxylin-eosin stain, original magnification ×60). [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

nerve paralysis, which recovered in the following 4 months. The patient is alive and disease free 3 years after the initial diagnosis.

DISCUSSION

Mucoepidermoid carcinoma is an epithelial tumor and the most common salivary gland malignancy. Its presence in Stensen's duct is extremely rare, which led us to review the literature to discuss its incidence, diagnosis, treatment, and prognosis. A Medline search with the terms "Stensen's duct and malignant tumors" revealed 30 reported cases.[2–4,6–25] After critical and meticulous review, we excluded six of these 30 reported cases, because they involved tumors that were not primary to Stensen's duct but that originated from the parotid gland or from the accessory parotid gland with secondary duct involvement.[6–9] The 24 remaining cases involved primary malignant tumors of Stensen's duct, and these tumors were of the following four different histologic types: squamous cell carcinoma (n = 8),[7,11,13–18] mucoepidermoid carcinoma (n = 5),[2–4] adenocarcinoma (n = 4),[19–21] and various types (n = 7; adenoid cystic carcinoma [n = 3],[3,12,25] sarcoma [n = 2],[22,24] undifferentiated carcinoma [n = 1],[23] and salivary duct carcinoma [n = 1][10]).

Including the case described herein, six cases of mucoepidermoid carcinoma of Stensen's duct have been reported. The five previously reported cases were by the following authors: Gaisford et al[2] (three cases); Clairmont et al[3] (one case); and Haar et al[4] (one case). The clinical data are summarized in Table 1. The patients' ages ranged from 25 to 61 years (mean, 52 years). The mucoepidermoid carcinoma of Stensen's duct has a female predilection, similar to its incidence in the parotid gland, with a female/male ratio of 2:1. Physical symptoms depend on the size, rate of growth, and delay to tumor diagnosis. Reported symptoms include cheek swelling, palpable mass, recurrent parotitis, or paralysis of the facial nerve. Grading is available in only three reported cases, with one each being high, intermediate, and low grade (Table 1). The tumor size (T classification) was available in all cases; four cases involved T1 tumors and two involved T3 tumors (Table 1). The small tumor size at the time of diagnosis is probably caused by an obstruction of Stensen's duct followed by early symptoms. In addition, a lump in the cheek is more easily detected by the patient than are other lesions and is not surrounded by much soft tissue, such as lesions within the parotid gland.

Abnormalities of Stensen's duct can be explored by use of several techniques. Besides traditional sialography, sonography, CT scan, and MRI, new techniques such as MR sialography[26,27] and sialoendoscopy[28] are now available. By providing a preoperative diagnosis, those techniques may help in treatment planning. Radiographic sialography is traditionally the standard for exploring abnormalities of the salivary ducts.[29,30] MR sialography with heavily T2-weighted images is considered a new and promising technique that will probably replace standard sialography.[26,27] The most recent diagnostic and interventional technique is sialoendoscopy, which is probably one of the most reliable procedures to provide information on salivary duct diseases.[28] The sialendoscope with a working channel for instrumentation permits the introduction of a biopsy forceps to obtain tumor tissue for preoperative diagnosis. In our case, Stensen's duct obstruction was observed by use of the sialoendoscopy. With the newly developed instruments, this minimally invasive technique could help in preoperative diagnosis and in deciding consequent treatment strategies.[28] Unfortunately, in our case, the programmed interventional sialoendoscopy could not be performed because of lack of patient compliance.

In general, treatment of patients with mucoepidermoid carcinoma of the parotid gland depends on the tumor grade, clinical stage, and adequacy of surgical treatment. Does mucoepidermoid

Table 1. Case reports of mucoepidermoid carcinoma of Stensen's duct.

Author, y	Patient age, y/sex	Symptoms	Grade	Stage	Treatment	Outcome
Gaisford et al, 1965[2]	55 / F	Large, ulcerated, bleeding mass left bucal area	NS	T3N?M?	Total parotidectomy, resection of the inner aspect of the cheek, radical neck dissection	Immediate recurrence, death followed promptly
	25 / F	Firm mass right bucal area	NS	T1N?M?	Superficial parotidectomy, excision of Stensen's duct, radiation therapy (due to incomplete surgery)	No recurrence for 18 mo
	NS / M	Cystic mass in the center of right cheek area	NS	T3N?M?	Total parotidectomy, extensive dissection of cheek and mouth	NS
Clairmont et al, 1979[3]	53 / F	Firm, mobile, nonpainful, 1- × 1-cm mass of the right cheek area	Intermediate	T1N0M0	Total parotidectomy, excision of Stensen's duct	No recurrence, follow-up NS
Haar et al, 1991[4]	57 / M	1-cm mass anterior to the left parotid gland region	High	T1N0M0	Superficial parotidectomy, excision of Stensen's duct	No recurrence for 6 y
Present case	61 / F	1.5-cm subcutaneous mass located below the left zygomatic arch	Low	T1N0M0	Superficial parotidectomy, excision of portion of the masseter muscle and of the Stensen's duct	No recurrence for 3 y

Abbreviations: F, Female; NS, not stated; M, male.

carcinoma of the Stensen's duct deserve a different treatment approach than its parotid gland counterpart? Because of the rarity of this disease, the answer may not be clear. However, in the reported cases, treatment seemed to take into consideration the clinical stage and the resection margins. It varied from total parotidectomy, neck dissection, and extensive dissection of the cheek and mouth to partial parotidectomy and excision of the Stensen's duct followed by radiotherapy in cases in which the resection was incomplete. Thus, treatment approaches were similar to those seen in mucoepidermoid carcinoma of the parotid gland itself. Because malignant parotid duct tumors are rare, we would rather follow the general guidelines for treatment of malignant parotid gland tumors. Treatment guidelines broadly depend on mucoepidermoid carcinoma grade, extent of surgery, facial nerve preservation, neck dissection, and the role of postoperative radiotherapy.[31]

Patient outcome was cited in six cases, including ours. Four of the five patients with stated follow-up had a good prognosis, with no recurrence after a follow-up of 1.5 to 6 years. The outcome seemed to be related to the clinical stage, because all four patients without recurrence had an early-stage tumor (T1). However, because of the small number of cases, we cannot draw any precise conclusions in this regard.

In summary, we presented a case of a mucoepidermoid carcinoma arising in Stensen's duct in a 61-year-old woman treated with superficial parotidectomy and excision of the Stensen's duct, without additional therapy. The patient was alive and free of disease 3 years after surgery. We also summarized the clinical presentation, diagnosis, therapy, and prognosis of the previously reported cases of mucoepidermoid carcinoma of Stensen's duct. The new available imaging techniques (MR sialography, sialoendoscopy) could be used to help in the diagnosis and in planning patient management. Mucoepidermoid carcinoma of Stensen's duct should be treated like the mucoepidermoid carcinoma occurring in the parotid gland, taking into consideration the clinical stage, tumor grade, and excision margins.

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