Available online at www.sciencedirect.com

ScienceDirect

British Journal of Oral and Maxillofacial Surgery 57 (2019) 365–367

BRITISH Journal of Oral and Maxillofacial Surgery

www.bjoms.com

Short communication

Lymphoepithelial carcinoma of the accessory parotid gland

C.A. Salcedo-Gil [a,*], A. Prieto Gundin [b], M. Alberola Ferranti [c], J. Rubio-Palau [d]

[a] *Division of Maxillofacial Surgery, Department of Pediatric Surgery, Hospital Sant Joan de Deu (Barcelona Children's Hospital), Esplugues de Llobregat, Barcelona, Spain*

[b] *Department of Pediatric Anesthesiology, Hospital Sant Joan de Deu, Esplugues de Llobregat, Barcelona, Spain*

[c] *Department of Pathology, Hospital Vall d'Hebron, Barcelona, Spain*

[d] *Division of Maxillofacial Surgery, Department of Pediatric Surgery, Hospital Sant Joan de Deu (Barcelona Children's Hospital), Esplugues de Llobregat, Barcelona, Spain*

Accepted 27 September 2018
Available online 28 March 2019

Abstract

Lymphoepithelial carcinoma of the accessory parotid gland is rare, and to our knowledge, only two cases have previously been reported. It has an association with the Epstein-Barr virus and is usually seen in Asians and Greenland Eskimos. We report a case of lymphoepithelial carcinoma of the left accessory parotid gland in a 59-year-old European man who had been raised in the Belgian Congo. After excision of the left accessory parotid gland with preservation of the facial nerve, he recovered well without complication, and there was no evidence of locoregional recurrence or distant metastases after follow up of 3.5 years.

Keywords: Lymphoepithelial carcinoma; Parotid tumours; Accessory parotid gland

Introduction

Lymphoepithelial carcinoma was first described by Hilderman et al in 1962.[1] It is rare, accounting for less than 1% of all tumours of the major salivary glands, and to our knowledge, has been described in the accessory parotid gland only twice before.[2]

Case report

In June 2014, a 59-year-old man presented at a regular dental check with a non-tender, painless mass in the centre of his left cheek. He had no history of exposure to radiation, facial weakness, or cervical lymphadenopathy, but had lived during his childhood in the Belgian Congo where there is a high incidence of lymphoepithelial carcinoma and Epstein Barr virus.

Physical examination showed a 1 cm firm, mobile, mass (Fig. 1) with no sign of lymphadenopathy or involvement of the facial nerve.

Magnetic resonance imaging (MRI) showed a nodular mass with a maximum anteroposterior diameter of about 15 mm in the left hemifacial region outside the masseter muscle and aligned with Stensen's duct, which suggested pleomorphic adenoma of the accessory parotid gland (Fig. 2). Fine-needle aspiration biopsy specimens failed to give a definitive diagnosis.

The patient had excision of the left accessory parotid gland with preservation of the facial nerve (Fig. 3). Histopathological analysis showed a high-grade, poorly-differentiated

* Corresponding author. Present address: Passeig de Sant Joan de Déu, 2, 08950, Barcelona, Spain.
E-mail address: carlosa.salcedo@ssib.es (C.A. Salcedo-Gil).

https://doi.org/10.1016/j.bjoms.2018.09.017

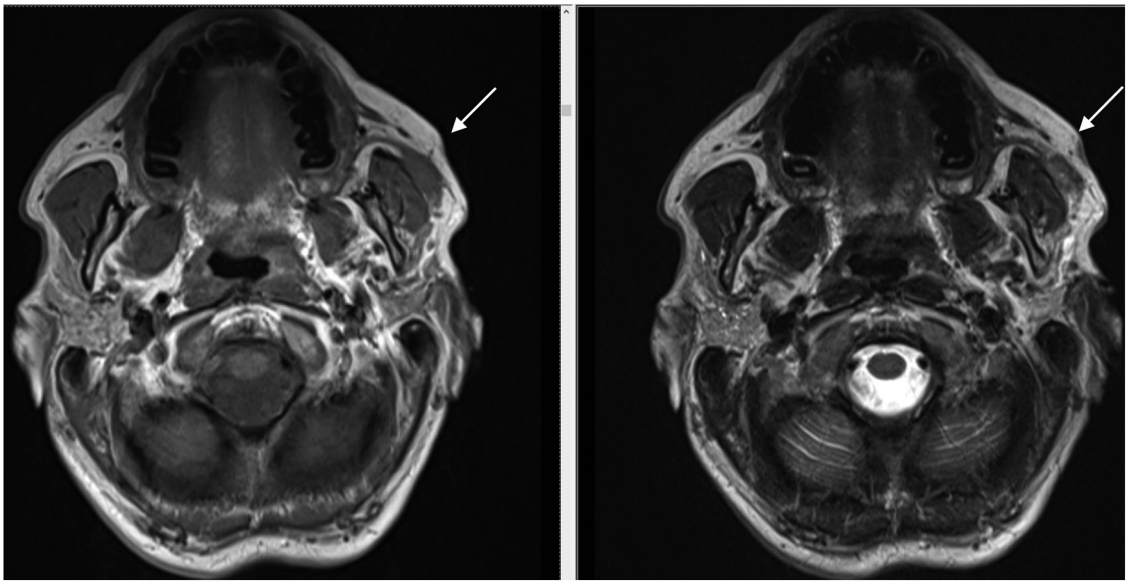

Fig. 1. Axial view of a magnetic resonance image showing a nodular mass in the left hemifacial region outside the masseter muscle and aligned with Stensen's duct.

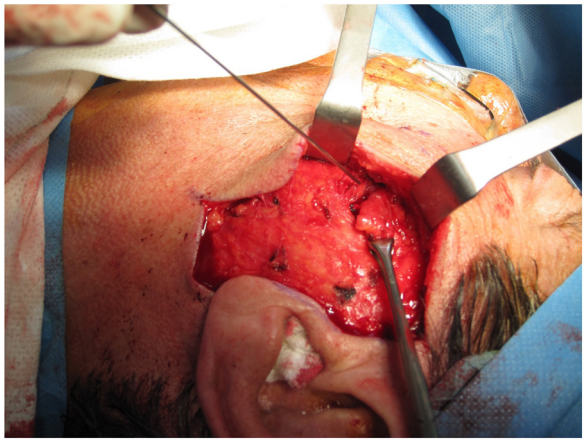

Fig. 2. Preoperative photograph showing the mid-cheek mass.

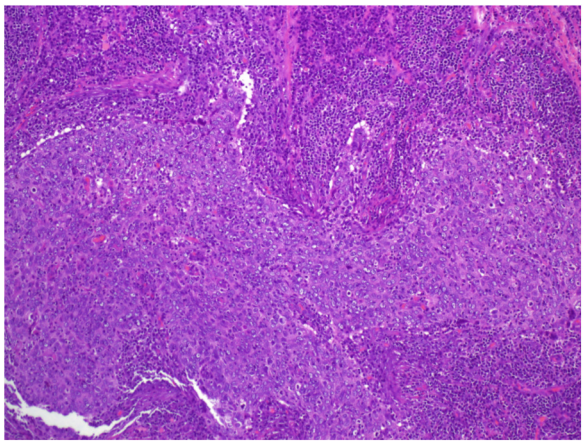

Fig. 3. High-grade, poorly-differentiated lymphoepithelial carcinoma, with clear margins (haematoxylin and eosin, original magnification x 10). Fibroadipose tissue showing a marked lymphoplasmocytic component with the formation of lymphoid aggregates. Atypical epithelial cellularity forms solid nests. Cytoplasms are broad, with eosinophils and microvacuoles. The nuclei are oval with a reinforced membrane and eosinophilic nucleoli. Apoptosis and abundant mitoses can be seen. The widened excretory duct is surrounded by a lymphocytic component but no residual parotid gland can be seen.

lymphoepithelial carcinoma with clear margins (Fig. 4). Epstein-Barr virus and p16 were not found.

Positron emission tomography (PET) showed no distant metastases. A detailed examination of the nasopharynx and Waldeyer's ring (which included fibreoptic nasopharyngoscopy) found no lesions, so no biopsies were taken.

Clinical follow up and a PET computed tomogram (CT) at three months postoperatively showed no evidence of locoregional recurrence or distant metastases, so neck dissection and postoperative radiotherapy were deemed unnecessary. At the 3.5-year follow up, the patient was free from disease.

Discussion

The accessory parotid gland has been described as salivary tissue that is anterior to, and separate from, the main body of the parotid gland, and adjacent to Stensen's duct. Tumours here are rare. They account for between 1% and 7.7%[1,2] of all parotid gland tumours, and between 26% and 50% of them are malignant.[3]

Lymphoepithelial carcinoma of the salivary glands is a rare, malignant salivary gland tumour that is associated with undifferentiated carcinoma and shows interstitial infiltrations by lymphocytes and plasma cells.[1,2,4,5]

Epidemiologically, it has an obvious geographical and racial predisposition, and mainly affects Eskimos, Green-

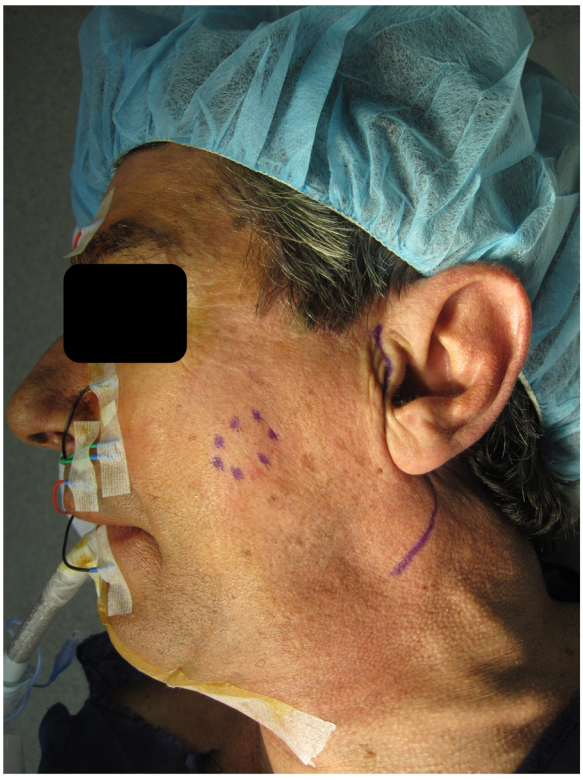

Fig. 4. Excision of the accessory parotid gland with preservation of the facial nerve by nerve monitoring.

landic Inuit, the Japanese, and people from the southern coastal region of China.[1,2,5–7]

Lymphoepithelial carcinoma of the salivary glands accounts for only 0.4% of all salivary gland tumours and has close associations with the Epstein-Barr virus.[2,4,7] Serological testing has shown that this is so in nearly all endemic, and most non-endemic cases.[2] The exact origin and pathogenesis of parotid lymphoepithelial carcinoma remains unknown.[2,5,6]

All patients should have a careful physical examination of the head and neck. Additional diagnostic tools include fine-needle aspiration, CT, and MRI.[2,3] The possibility of nasopharyngeal carcinoma must be excluded by clinical examination of the upper respiratory and digestive tracts before a diagnosis of lymphoepithelial carcinoma can be accepted.[1,2,5,6]

The cervical lymph nodes are involved in up to 40% of patients, and 20% develop distant metastases within three years of being treated.[4,5] The disease metastasses most commonly to the lungs, liver, brain, and bone.[5,6] Rates for local, regional, and distant metastases, are high because lymphoepithelial carcinoma is similar to nasopharyngeal carcinoma in histocytology and behaviour.[1,2]

Salvage surgery and chemotherapy are recommended in patients with local or regional recurrence. The disease is a high-grade malignancy, and five-year survival has been reported to range from 70% to 85%.[5,6]

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patient's permission

The authors have obtained the patient's informed consent. The document is held by the corresponding author.

References

1. Hsu YC, Lu HF, Huang CC, et al. Malignant lymphoepithelial lesions of the salivary gland. *Otolaryngol Head Neck Surg* 2006;**134**:661–6.
2. Manganaris A, Patakiouta F, Xirou P, et al. Lymphoepithelial carcinoma of the parotid gland: is an association with Epstein-Barr virus possible in non-endemic areas? *Int J Oral Maxillofac Surg* 2007;**36**:556–9.
3. Lukšić P, Suton M, Rogić L, et al. Accessory parotid gland tumours: 24 years of clinical experience. *Int J Oral Maxillofac Surg* 2012;**41**:1453–7.
4. Roy, Moubayed SP, Ayad T. Lymphoepithelial carcinoma of the sublingual gland: case report and review of the literature. *J Oral Maxillofac Surg* 2015;**73**:1878. e1–5.
5. Santamaría AT, Dasi BL, Moleón LG, et al. Primary lymphoepithelial carcinoma of the parotid gland. *Revista Española de Cirugía Oral y Maxilofacial* 2013;**35**:96–9 (paper in Spanish).
6. Wu DL, Shemen L, Brady T, et al. Malignant lymphoepithelial lesion of the parotid gland: a case report and review of the literature. *Ear Nose Throat J* 2001;**80**:803–6.
7. Christiansen MS, Mourad WA, Hales ML, et al. Spindle cell malignant lymphoepithelial lesion of the parotid gland: clinical, light microscopic, ultrastructural, and in situ hybridization findings in one case. *Mod Pathol* 1995;**8**:711–5.