American Journal of Otolaryngology and Head and Neck Surgery

Case Report
Published: 21 Jun, 2018

Reconstructive Surgery in Adenoid Cystic Carcinoma of Minor Salivary Glands of Oral Mouth: Our Experience

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Abstract

Adenoid cystic carcinoma is an uncommon salivary gland tumour that often may arise with an advanced stage at diagnosis. The clinical and pathological patterns are characterized by slow growth, peri-neural invasion, multiple local recurrences and distant metastases. The optimal treatment is generally radical surgical resection and is almost always followed by postoperative radiotherapy. Much effort has been invested into understanding the tumour's molecular biological processes, aiming to identify patients at high risk of recurrence, in hope that they could benefit from other, still unproven treatment modalities such as chemotherapy or biological therapy. This article report two cases of adenoid cystic carcinoma of oral anterior floor treated in our Department of Otorhinolarygology of Tor Vergata University Hospital, analysing each one clinical presentation, radiological diagnosis, surgical reconstructive treatment, immuno-histopathological pattern, clinical and radiological follow up and overall survival.

Keywords: Adenoid cystic carcinoma; Oral floor; Minor salivary glands; Reconstructive surgery

Introduction

In 1853 Robin, Lorain and Laboulbene first described two cases of an uncommon epithelial tumour of the nose and the parotid gland, which was named "cylindroma" by Billroth in 1856 [1,2]. Only in 1930 Spies introduced the term "Adenoid Cystic Carcinoma" (AdCC), and until 1940s AdCC was considered a benign variant of the mixed salivary gland tumour [3]. The malignant nature of this neoplasm was finally explained by Dockerty and Mayo in 1943 [4].

AdCC represents about 10% of salivary gland tumours [5] and about 1% of all head and neck malignant neoplasms [6]. Although AdCC is rare, it can be considered the most common malignant neoplasm of the submandibular and minor salivary glands but it can also occur in different sites of head and neck regions where secretory glands are, such as nose and paranasal sinuses, trachea, larynx and even lacrimal and ceruminous glands [6-10].

AdCC frequently appears as a small, slow growing, lesion, but it is often discovered at an advanced stage [11]. The main characteristics of this type of neoplasm are peri-neural invasion, which occurs in around 22% to 46% of cases, and multiple local recurrencies. Regional lymph-node involvement is considered rare. However, distant metastases have a 40% incidence, with lung, bone, and liver representing the most commonly affected sites [3]. Clinically, pain is the main symptom. The treatment of choice is represented by radical surgical resection, often followed by post-operative radiation therapy and, in selected cases, by chemotherapy [7]. Minor salivary gland AdCCs seem to have a worse prognosis than the major salivary glands' ones. In most cases, this neoplasm has a long course and uncertain prognosis. It has been observed that some asymptomatic patients affected by advanced and unresectable AdCCs who were not treated, as well as patients with stable metastatic disease, may survive even for 10 years to 15 years.

Case Report

Case 1: A 49-year-old man affected for almost 15 years by an asymptomatic right submandibular lesion, which grew in the last years, referred to our Unit and performed a ^{18}FDG PET-CT scan which showed an increase of the metabolic activity of the submandibular lesion as well as a hyper fixation of the metabolic tracer in a nodular formation of about 20 mm in the apical segment of the upper lobe of the left lung, with irregular contours (Figure 1A). The needle-biopsy of lesion was positive for adenoid cystic carcinoma. The patient underwent first video-assisted thoracoscopic typical resection of the upper lobe of left lung and then a right emi-mandibulectomy, bilateral functional neck dissection, reconstruction with a mandibular plaque and myocutaneuos flap with right

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Received Date: *21 May 2018*
Accepted Date: *18 Jun 2018*
Published Date: *21 Jun 2018*

Citation:
De Berardinis R. Reconstructive Surgery in Adenoid Cystic Carcinoma of Minor Salivary Glands of Oral Mouth: Our Experience. Am J Otolaryngol Head Neck Surg. 2018; 1(3): 1013.

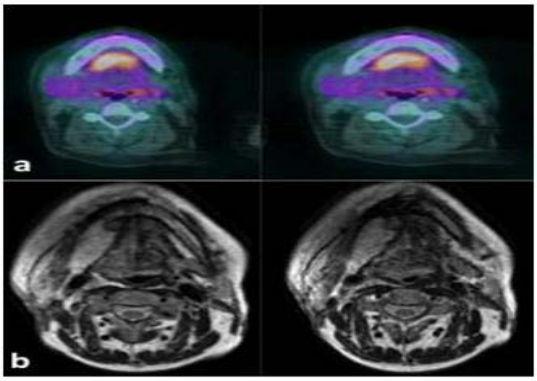

Figure 1: A) Pre-operative PET-CT axial scan; B) Post-operative MRI axial sc.

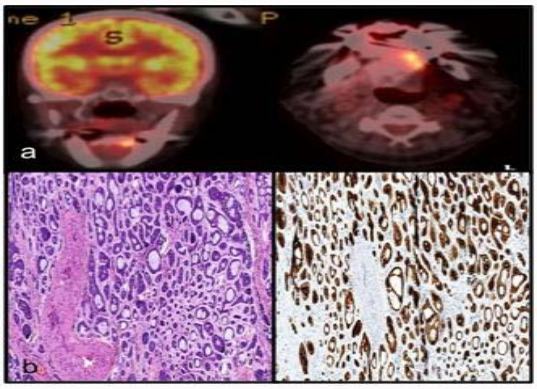

Figure 2: A) Pre-operative PET-CT scan; B) Histological images: on the left tubular pattern displaying tumour cells surrounding pseudocystic spaces, containing amorphous material; on the right reactivity of adenoid cystic carcinomas for CK7.

pectoralis major muscle, temporary tracheotomy. Histological report showed a neoplasm with common aspects of peri-neural infiltration, solid pattern of growth (grade III) and invasion of surrounding parenchyma; 4 of 18 lymph-nodes were neoplastic. Surgical margins were negative for neoplastic infiltration (R0). Immunohistochemical pattern of the lesion was characterized by positivity for CD14, panCK, CK7, EMA and c-Kit, S100, p63 focally; proliferative index was 40% of neoplastic cells. The left lung lesion showed to be a metastatic localization of the disease, with a proliferative index of 45% and negativity for c-Kit. Nutritional therapy was guaranteed by nasogastric tube that was removed on 10th post-operative day, starting a liquid diet. Outcomes of surgery were good, except for a deficit of the marginalis mandibulae branch of right facial nerve (Figure 1B). No adjuvant therapy was performed. The current survival is about 24 months after treatment and the patient is still alive without the disease.

Case 2: A 66-year-old woman referred to our Unit due to the occurrence of left tongue paresthesia and tenderness in the left sublingual space in the last two months. Ultrasonography examination revealed a hypoechoic solid nodule of about 14 × 11 mm on the left portion of the mouth floor and presence of inner vascularization at the color- and power-Doppler evaluation. MRI showed a moderately and heterogeneously hyperintense lesion on T2-weighted sequences of 24 × 14 mm, with irregular margins, infiltrating the genioglossus and mylohyoid muscles. Before radical surgery, ^{18}FDG PET-CT was performed to stage the disease (Figure 2A); there were no evidences of metastatic disease. The patient underwent excision biopsy and pathology on the specimen showed an adenoid cystic carcinoma of minor salivary gland. Therefore, an *en-bloc* excision of the floor of the mouth with "pull-through" technique was performed, with reconstruction with skin-flap of infra-hyoid muscles and temporary tracheotomy. Definitive histologic report described a massive peri-neural infiltration of surrounding tissue; immunohistochemical pattern showed positivity for panCK, CK7, c-Kit, p63, CD14 (Figure 2B), and proliferative index with Ki67 equal to 20% of neoplastic cells. Nasogastric tube allowed nutritional therapy. Surgical margins were negative for neoplastic infiltration (R0). No post-operative complications occurred and no adjuvant therapy was performed. Through rehabilitation therapy, the patient has recovered a good speech and an efficient oral phase of swallowing, ensured by the reconstruction with myocutaneous flap. Overall survival was of about 12 months and patient is still alive.

Discussion

Adenoid Cystic Carcinoma (AdCC) represents about 10% of salivary gland tumours and about 1% of all head and neck malignant neoplasms [1]. Although it is rare, it can be considered the most common malignant neoplasm of the submandibular and minor salivary glands but it can also occur in different sites of head and neck regions [2,3].

AdCC frequently appears as a small, slow growing, lesion, but it is often discovered at an advanced stage and generally pain is the main symptom [4]. The main characteristics of this type of neoplasm are peri-neural invasion, which occurs in around 22% to 46% of cases, and multiple local recurrencies [5], while distant metastases have a 40% incidence [6]. Minor salivary gland AdCCs and solid pattern seem to have a worse prognosis [7].

Radical surgery with wide resection margins represents the treatment of choice in AdCC, often followed by post-operative radiation therapy and, in selected cases, by chemotherapy [8]. The goal of obtaining disease-free margins is generally not achieved due to both AdCC's frequent tendency to peri-neural invasion and the challenging anatomical access some lesions can present with. In particular, the surgical treatment of minor salivary glands AdCC depends on the site of origin and on the extent of the tumour and in many cases surgical reconstruction with flap is required [9].

Post-operative radiation therapy is commonly performed in patients with regression and/or relief of symptoms, with peri-neural invasion, neck lymph-nodes metastases, recurrent tumours and solid histological subtype and in all cases of unresectable tumours or as symptomatic palliation [10]. In addition to traditional radiation therapy with photons, therapy with carbon ions that seems to increase local disease control to 82% at 5 years in combined therapies with IMRT (Intensity Modulation Radiation Therapy) [10].

Lymph-node metastases are only occasionally seen in AdCC but elective neck dissection is generally recommended by most Authors for staging and achieving regional control of the disease [8]. Systemic chemotherapy represents a controversial treatment, due to a reported low sensitivity of AdCCs to this kind of treatment; however, palliative chemotherapeutic treatment proved to be useful in a small percentage of patients with recurrent or metastatic disease [11].

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