Oral Oncology 92 (2019) 95–98

Contents lists available at ScienceDirect

Oral Oncology

journal homepage: www.elsevier.com/locate/oraloncology

Letter to the editor

Ossified Carcinoma Ex Pleomorphic Adenoma in accessory lobe of parotid gland: Complexity in clinical, imaging and histologic diagnosis and minimally invasive surgery

ARTICLE INFO

Keywords:
Carcinoma ex pleomorphic adenoma
Accessory lobe of parotid gland
Ossified carcinoma
Head and neck neoplasm
Minimally invasive surgery

ABSTRACT

Carcinoma ex pleomorphic adenoma is not common among malignant salivary tumors and an accessory lobe of parotid gland tumor with a carcinoma ex pleomorphic adenoma histology is even rarer. Management of these tumors include a high index of suspicion, good understanding of the anatomy and a meticulous surgical access. The exceptional localization of an ossified Carcinoma ex Pleomorphic Adenoma in the accessory lobe of the parotid gland, never reported to date in the literature, has prompted us to report this case to stress the challenging difficulties related to clinical and histologic diagnosis and to analyze a minimally invasive intraoral approach.

Introduction

Accessory lobe of parotid gland (APG) is present in approximately 21% of the population and tumors in this location are 1% of all parotid tumors [1,2], thus a mass arising in the midcheek region may often be overlooked as an accessory lobe of parotid neoplasia. APG is associated with a higher rate of malignant tumors (26–50%) than main parotid gland (18–20%) [3,4]. Carcinoma ex pleomorphic adenoma (CEPA) is not common among malignant salivary tumors (4.3–7.8% of malignant parotid tumors) and its localization in APG is even rarer (1%) [5,6]. Ossification in parotid CEPA is very rare [7,8], and to the best of our knowledge, no case of ossified carcinoma ex pleomorphic adenoma arising in the accessory lobe of parotid gland has been reported in literature.

The exceptional localization of ossified CEPA in the accessory lobe of the parotid gland has prompted us to report this case to stress the challenging difficulties related to clinical and histologic diagnosis and to analyze a minimally invasive intraoral approach (Fig.1).

Case report

A 50 years old man was referred to Cranio-Maxillo-Facial Surgery Department, University of Campania, Naples. The patient had a medical history of Non-Hodgkin Lymphoma treated with radiochemoterapy and autologous bone transplant. Physical examination revealed a painless, 1.5 cm, barely defined elastic smooth surfaced midcheek mass, been present 11 months, with recent increase in size. The skin overlying the mass was not hyperchromic or wrinkled. The mass, extending the thickness of the cheek, was equally appreciable on external and internal examination without abnormality of oral mucosa or salivary flow. No facial weakness or sensitivity alteration was reported. Laboratory examinations, including serum amylase levels, were within normal ranges. Ultrasonography and Computed Tomography revealed a circumscribed, homogeneously calcified, well-enhanced solitary mass, 31 × 12 mm in size, anterior to the front edge of the parotid gland, near the end of Stensen's duct and on the outer layer of the masseter muscle. No remarkable cervical lymphadenopathy was detected. A percutaneous fine needle aspiration cytology (FNAC) showed several epithelioid cells, organized in single elements and small clusters, characterized by evident nuclear atypia. Necrotic material was observed in the background. Surgery was performed using an intraoral approach and a clearly defined dissection plane was not found. The tumor was detected in the contest of the accessory lobe and it was removed en-bloc together with the surrounding tissues. Tumor invasion into the contiguous salivary tissue was not observed. The parotid duct and facial nerve branches were successfully preserved: neither facial paralysis nor salivary fistula was observed postoperatively. Histological examination confirmed the intraoperative evidence of an ossified mass: osseous mineralized tissue was observed when tissue slices were made. Microscopic examination showed a glandular-patterned cellular proliferation with abundant hyaline, calcified and ossified stroma. The neoplasia was partially delimitated by a fibrous capsule, but infiltration of the adjacent tissues was focally observed. In correspondence of the infiltrative areas, the neoplastic cells showed moderate nuclear atypia. Immunoistochemical evaluations demonstrated a double cellular population: luminal cells resulted positive for cytokeratin CK 7 and CK19, while basal cells resulted positive for Glial fibrillary acidic protein (GFAP) and S100. In malignant areas, intense nuclear immunoreactivity for Ki-67 was presented. Smooth muscle actin and CD117 resulted negative. A final diagnosis of high-grade CEPA was rendered (Fig. 2). The patient was followed up closely: after oncologic consulting, radiation therapy was recommended.

https://doi.org/10.1016/j.oraloncology.2019.03.003
Received 25 February 2019; Accepted 4 March 2019
Available online 07 March 2019
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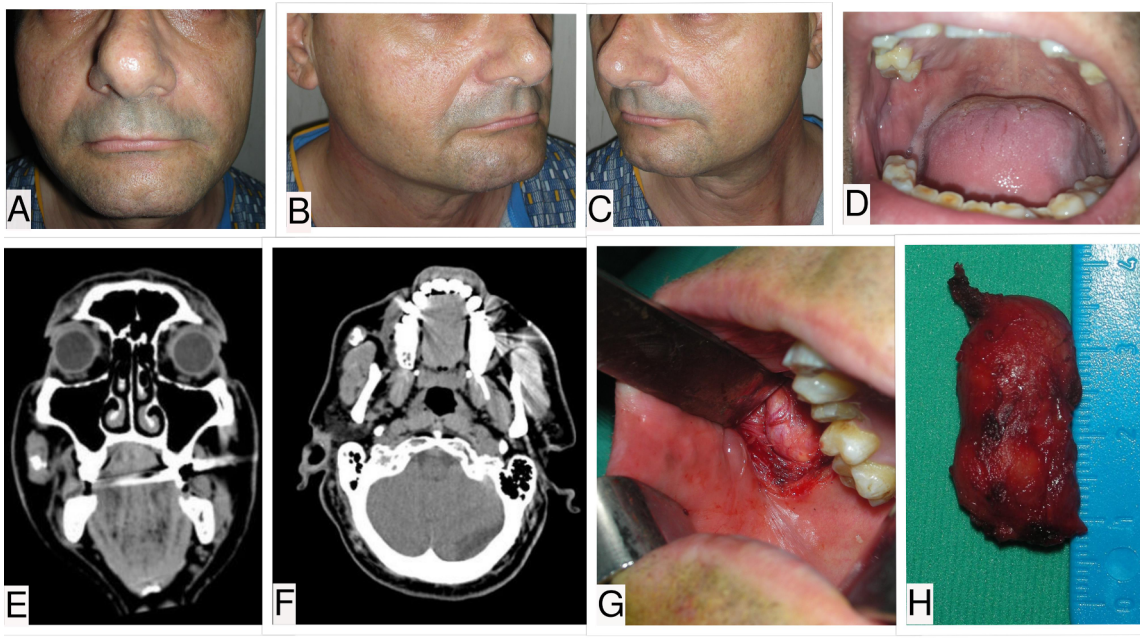

Fig. 1. A-B-C: Frontal (a), three-quarters right (b) and three-quarter left (c) view of a 55 years old male patient presenting right midcheek swelling. **D:** Intraoral examination in the right side showing swelling without noticeable abnormality of mucosa or salivary flow. **E-F:** CT imaging showing circumscribed, homogeneously calcified, well-enhanced solitary mass, 31 × 12 mm in size, anterior to the front edge of the parotid gland, near the end of Stensen's duct and on the outer layer of the masseter muscle. **G:** Intraoperative picture: intraoral surgical approach from intraoral view. Note facial nerve branches running over the tumor surface. **H:** Operative specimen. The final histopathologic diagnosis was accessory lobe Carcinoma Ex Pleomorphic Adenoma.

Discussion

CEPA represent the 4% of all salivary neoplasia. It is an aggressive tumor with high overall mortality, male predilection (M: F = 2: 1) and occurs at the age from 35 to 90 years (mean 62 years) [9,10]. At the clinical evidence, the volume is often small (mean size 3.9 cm): symptomatic mass is the most common symptoms that could lead to a tardive diagnosis. The importance of an early diagnosis is related of the date that survival is largely related to clinical staging: 5-year survival for stage I is over 80%, the overall5-year survival decrease to 37%. Malignant transformation of pleomorphic adenoma in CEPA increases with the duration of the tumor (1.6% in 5 years Vs 9.5% in 15 years) [11,12] and p53loss, p16deregulation and PLAG1 and Ki-67 overexpression are involved. The role of radiation in the pathogenesis of CEPA remains speculative [13–16].

APG is located adjacent to Stensen's duct, separate from the main body of the parotid gland, between buccal and zygomatic facial nerve branches. The frequency of the presence of an APG is 20 of 96 cases [1,2]. Neoplasms of accessory lobe of the parotid are extremely rare, 1–7% of all parotid tumor, 35% of which are malignant tumors (CEPA has only the 1% of incidence):this could be attributable to APG histology, made up of an equal percentage of mucinous and serous acinar units [5,6].

Ossification of CEPA is extremely rare [7,8] and the diagnosis of calcified APG malignant tumor can be extremely challenging because of its confounding features. Because of their midcheek region localization, differential diagnosis is wide and includes Stensen's duct stone, soft tissue chondroma or osteoma, extracranial meningioma, vascular ossification of atherosclerotic lesions, Albright's syndrome and fibrodysplasia ossificans progressive. Thus, in the presence of a midcheck ossified mass is essential a high suspicion for malignancy although APG tumors are very rare [4–7].

In our case, extensive areas of ossification were merged in an abundant collagenous stroma and the presence of several cellular atipias, markers and infiltrative pattern of the stroma advocated the evidence of CEPA. Because of histological evidence of bone tissue within areas of collagenous tissue, we suggest enchondral ossification in CEPA. To the best of our knowledge, this is the unique case of ossified carcinoma ex pleomorphic adenoma in the accessory lobe of parotid gland.

In APG tumors surgical approaches, cheek incision is related to inadequate excision, consequent local recurrence and 40% of facial nerve damage [17,18] whereas facelift or parotidectomy incision allow good visualization of facial nerve [18]; an endoscopic technique could be adapted for small benign tumor [19]. In our opinion, an intraoral approach, barely described in literature [17,18–20], could be consider a safe and cosmetically appealing access in APG neoplasia because of the absence of skin incision or facial nerve damage.

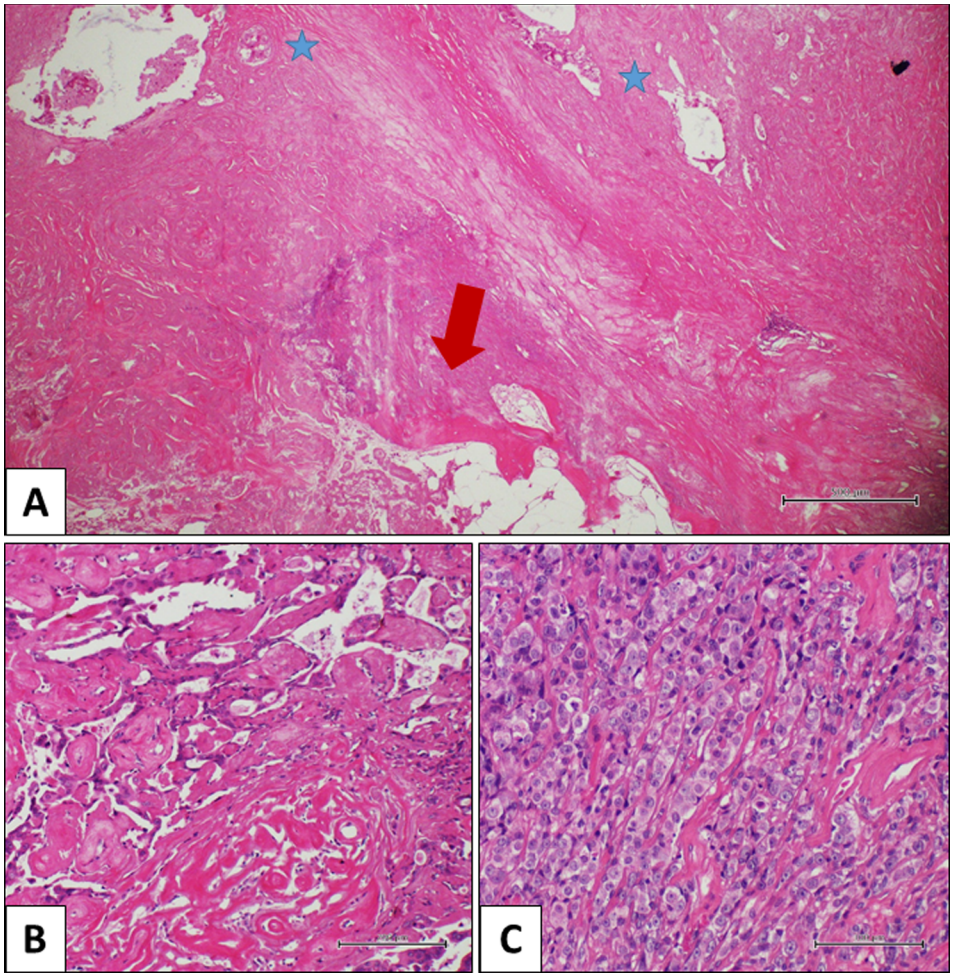

Fig. 2. Histological features. **A:** The neoplasm shows abundant collagenous stroma, with extensive areas of ossification (red arrow). The blue stars highlight neoplastic epithelial nests (H&E, 4x). **B:** Glandular elements with infiltrative pattern and hyaline stroma (H&E, 10x). **C:** In this field, the neoplasm shows a solid architectural pattern, with large epithelioid cells with atypical nuclei (H&E, 20x). H&E: hematoxylin and eosin.

Conflicts of interest

None declared.

Disclosures

None.

Funding

None.

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Raffaele Rauso[a], Giuseppe Colella[a], Renato Franco[b], Andrea Ronchi[b], Fabrizio Chirico[c,*]
[a] *Maxillofacial Surgery Unit, Department of Medical, Surgical and Dental Speciality, Campania University "Luigi Vanvitelli", Naples, Italy*
[b] *Pathology Unit, University of Campania "Luigi Vanvitelli", Naples, Italy*
[c] *Maxillofacial Surgery Unit, Department of Maxillo-Facial Surgery, University of Naples "Federico II", Naples, Italy*
E-mail address: fabriziochirico@hotmail.com (F. Chirico).

* Corresponding author at: Maxillofacial Surgery Unit, Department of Maxillo-Facial Surgery, University of Naples "Federico II", Via Sergio Pansini 5, 80131 Naples, Italy.