Oral and Maxillofacial Surgery Cases 6 (2020) 100188

Contents lists available at ScienceDirect

Oral and Maxillofacial Surgery Cases

journal homepage: www.oralandmaxillofacialsurgerycases.com

Adenoid cystic carcinoma of accessory parotid gland: A case report and review of the literature

Antonio Romano [a], Carmelo Lo Faro [a,*], Giorgio Iaconetta [b], Umberto Committeri [a], Giovanni Audino [a], Giovanni Salzano [a], Fabio Maglitto [a], Luigi Califano [a], Giovanni Dell'aversana Orabona [a]

[a] *Division of Maxillofacial Surgery, Department of Neurosciences, Reproductive and Odontostomatological Sciences, University of Naples "Federico II", Naples, Italy*

[b] *Division of Neurosurgery, University of Salerno, Salerno, Italy*

ARTICLE INFO

Keywords:
Adenoid cystic carcinoma
Accessory parotid gland
Salivary glands tumors
Malignancies
Mid-cheek mass
Surgical treatment

ABSTRACT

Accessory parotid gland (APG) is a small salivary gland tissue ranging from 0.5 to 1 cm diameter in size, located almost 7 mm anterior to the main parotid gland. APG is a common anatomical variant, but accessory parotid gland tumors are very rare, accounting for only 1%–7.7% of all parotid gland tumors. We present a case of adenoid cystic carcinoma involving accessory parotid gland, a very rare condition with only, to the best of our knowledge, other 7 cases described in literature. Surgical excision was performed and eventful postoperative recovery, without severe surgical complications and no signs of recurrence at 12 months of follow-up was obtained, with a good cosmetic result. An extensive review of the literature has been performed and the results are presented in order to establish a correct diagnostic-therapeutic protocol for these oncologic patients.

1. INTRODUCTION

Accessory parotid gland (APG) is a small salivary gland tissue ranging from 0.5 to 1 cm diameter in size, located almost 7 mm anterior to the main parotid gland. APG is a common anatomical variant, with a frequency in autopsy studies ranging from 21% to 56% [1,2].

APG tumors are very rare, accounting for only 1%–7.7% of all parotid gland tumors, with a malignancy rate ranging from 26% to 52% compared to the 18.5% malignancy rate for tumors of the main parotid gland [3,4]. According to the data extracted from the literature, adenoid cystic carcinoma in the accessory parotid glands has an incidence of 1.9% [5]. We report a case of a patient who presented to us with a mid-cheek swelling arising from the accessory parotid gland, and diagnosed as adenoid cystic carcinoma. To the best of our knowledge, this is the seventh case reported in literature.

2. PRESENTATION of case

In November 2018, a 52-year-old man was presented to Maxillo-Facial Unit of Federico II University, Naples, Italy with a painless

* Corresponding author.
E-mail addresses: carmelolofaro28@gmail.com, c.lofaro@studenti.unina.it (C. Lo Faro).

https://doi.org/10.1016/j.omsc.2020.100188
Received 30 January 2020; Received in revised form 5 September 2020; Accepted 7 September 2020
Available online 10 September 2020
2214-5419/© 2020 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

nodule in the right mid-cheek region. Accurate anamnesis revealed that the mass had been increasing slowly in size over the last 8 months. Initial imaging studies included ultrasound scanning, followed by fine-needle aspiration cytology (FNAC) biopsy and CT scans. On clinical examination he presented an oval, firm and slightly movable, subcutaneous mass in the right mid-cheek region, measuring 2,5 × 1,5 cm with elastic consistency (Fig. 1). All cranial nerves were intact and no cervical adenopathy was palpated. Ultrasound scanning revealed a hypodense mass of 23 × 14 mm in size located under the skin, around the anterior edge of the masseter muscle, originating from the right accessory parotid gland. The patient underwent a first fine needle aspiration cytology (FNAC) examination that showed an inconclusive diagnosis, and a second one that instead showed "mucoid material with epithelial elements through acinar cell aggregates without atypic cells compatible with adenomatous proliferation". The computed tomography scans (Fig. 2A and B) confirmed a cystic nodule anterior to the parotid gland, non-vascularized, measuring 22 × 12 × 15 mm, with no pathologic cervical lymphadenopathy.

The patient underwent a superficial parotidectomy. The lesion was handled through a "facelift" incision, achieving a good exposure for the accessory lobe. The tumor was dissected out completely, together with superficial lobe of parotid gland (Fig. 3). The branches of facial nerve were not involved by the tumor, and the facial nerve monitor (Nerve Integrity Monitor, Medtronic, Minneapolis, MN) was used intra-operatively to facilitate nerve identification and minimize the risk of nerve injury. Postoperatively, transient facial nerve weakness (Grade II House-Brackman Grading System) was observed and it resolved after 3 months. On final histopathologic analysis, diagnosis consisted of cribriform adenoid cystic carcinoma (Fig. 4), surgical margins were negative for tumor involvement, with no perineural infiltration. The CT scan follow-up was performed 6 months after surgery, and it confirmed the total removal of the lesion.

The final staging was Stage 1 (T2 N0 M0) and the follow-up period is currently 12 months after surgery. There is no evidence of recurrence and no other complications, including facial nerve or auricular nerve permanent injury, salivary fistula, or Frey's syndrome, have been noted.

3. DISCUSSION

Accessory parotid gland is a small salivary gland tissue; it varies in size ranging from 0.5 to 1 cm and is located almost 7 mm anterior to the main parotid gland, between the zygomatic arch and the Stensen's duct, lying in continuity with the masseteric muscle, along an imaginary line that extends from the tragus to a midway point between the nose wing and the vermilion border of the lip [6,7]. APG has its own blood supply arising from the transverse cervical artery, and has an own duct emptying into Stensen's duct. Approximately 60% of benign tumors of the mid-cheek region originate from the accessory parotid glands [3]. Evaluation of a mass in the mid-cheek region can be challenging. Differential diagnoses of this area includes parotid gland cyst, accessory parotid gland tumors (APGTs) Arteriovenous malformation, neural tumors, adenopathy, and metastasis. APGTs should be suspected in any patient presenting with a midcheek mass [8], that most often present as indolent, progressively enlarging cheek mass [9]. Despite APG is a relatively common anatomical variant, whit a frequency in autopsy studies, from 21% to 56% [3,4]; APGTs are rare, accounting for only 1%–7.7% of all parotid gland tumors. Malignancy arises more often than malignancy of tumors of the main parotid gland, and the rate ranges from 26% to 52%. [7,8]. Treatment strategy consists in physical examination, ultrasonography, FNAC, followed by CT scans, and in uncertain cases MRI, in order to improve the accuracy of diagnosis [9]. Preoperative FNAC is a baseline investigative tool in the assessment of midcheek swelling [10] with a sensitivity in detection of malignant parotid gland tumors' raging from 60 to 73% [11]. We achieved a review of English-literature articles from the PubMed databases, regarding accessory parotid gland tumors' incidence and surgical treatment, using as key words "accessory parotid gland adenoid cystic carcinoma" and "accessory parotid gland tumors". A total of 58 papers focusing on APGTs have been identified, with two case reports of adenoid cystic carcinoma of the accessory parotid gland [12,13] and other four cases of adenoid cystic carcinoma included in larger series [3–5]. According to Dell'Aversana Orabona et al. [14], in the APG region, pleomorphic adenoma (PA) is the most common benign neoplasm. In the other hand, Luksic et al. [5] found that the most common malignant neoplasm is low-grade mucoepidermoid carcinoma with a malignancy

Fig. 1. Preoperative photographs: clinical examination shows swelling on the right mid-cheek region measuring 2,5 × 1,5 cm.

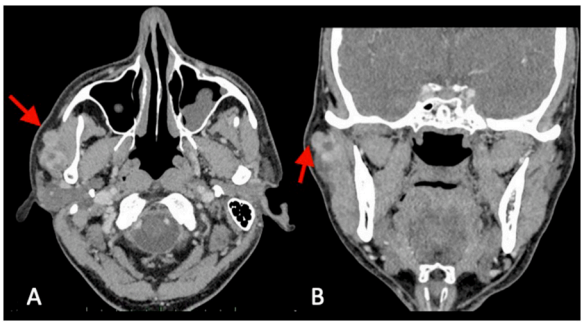

Fig. 2. Contrast-enhanced axial (A) and coronal(B) CT scans show cystic nodule anterior to the right parotid gland, non-vascularized, measuring 22 × 12 × 15 mm".

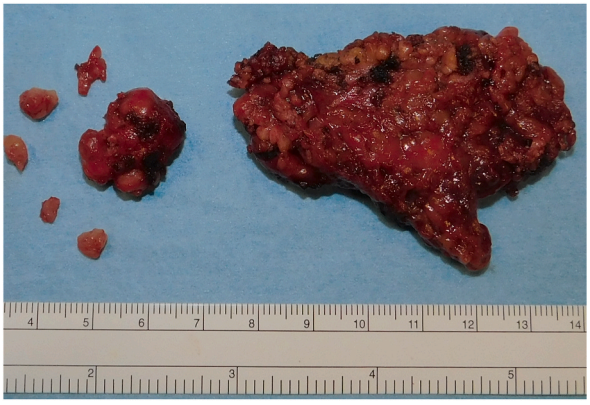

Fig. 3. Operative specimen: on the right the superficial parotid lobe, on the left the accessory parotid gland; final histopathologic diagnosis was cribiform adenoid cystic carcinoma.

rate of 10.2% (32/313) and that the adenoid cystic carcinoma has an incidence of 1.9% (6/313). These results are coherent with Rodino and Shaha [15] who found an incidence of 2% in a series of 150 cases, and slightly low if compared to the series of Yang et al. [3]. Eveson and Cawson [16] found that adenoid cystic carcinoma commonly appeared in the seventh decade of life with an average of 66.3 years, with a predominance in women (F:M = 1.2:1). Adenoid cystic carcinoma usually involves the parotid, submandibular and minor salivary glands and is well known for its perineural invasion. Clinically it appears as a slow-growing mass with local pain, facial paralysis due to neural invasion, and local invasion to deeper structures. Adenoid cystic carcinoma is characterized for three histopathological patterns: tubular, cribriform and solid. Huang et al. [17] observed a survival rate of 16.7% in cases with solid pattern, and 47.4% in cases with cribriform and tubular forms, 10 years after treatment. Batsakis et al. [18], further classified adenoid cystic carcinoma according to its histologic pattern and behavior into 3 grades. Grade I adenoid cystic carcinoma is a low-grade tumor with a 15-year survival rate of 39%, grade II has a 15-year survival rate of 20% and lastly grade III has only 5% of survival rate. Surgery is the basis of treatment for all grades of adenoid cystic carcinoma. The optimal surgical approach should provide safe access to the tumor, flexibility and easy manipulation with a satisfying aesthetic result. Radiotherapy (RT) or chemotherapy and RT also is used as an adjuvant therapy for intermediate and higher-grade tumors or tumors with adverse features such as positive margins and perineural invasion [19]. Surgical approaches described in literature, include direct skin incision overlying tumor [4–7], standard parotidectomy incision [2–11], facelift approach [8], intraoral excision [13] and endoscopic assisted resection with preauricular incision [14]. Our approach consists of a standard facelift incision, that allows exposure of the parotid gland and the accessory lobe, with a direct visualization of the tumor and the capability to identify and preserve distal nerve branches that may cross over the surface of the mass. We have found the nerve monitor to be a valuable tool to help in identifying small nerve branches. Complications include infection, local tumor recurrence, facial nerve paralysis, salivary fistula and Frey's syndrome [20]. Klotz and Coniglio [8] reported one case of salivary fistula in a series of four patients undergoing a standard parotidectomy approach for accessory parotid gland tumors, which resolved with antibiotics and local wound care. Johnson and Spiro [4] reported 4 facial nerve paralysis in a series of 10 patients undergoing a direct cheek approach compared with no facial nerve paralysis in 13 patients undergoing a standard parotidectomy approach. Perzik and White [1] reported no facial nerve injuries in 20 modified preauricular approaches. In our case there was no evidence of recurrence and no other complications, including infection, facial or auricular permanent injury, salivary fistula, or Frey's syndrome, were noted.

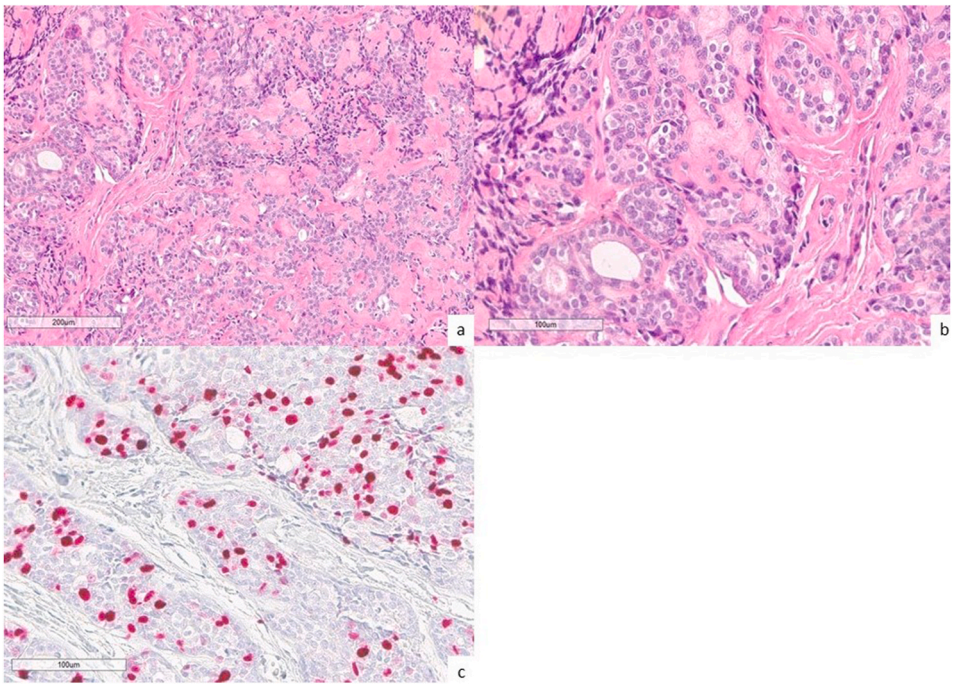

Fig. 4. Photograph of hematoxylin-eosin staining of the surgical specimen 10x (a) and 20x (b) show a tumor with tubular and diffuse pattern, focally cribriform, composed of basaloid appearance cells. Immunohistochemical staining (c) 20x, show expression for Ki67.

4. CONCLUSION

Adenoid cystic carcinoma of accessory parotid gland is a very rare condition. To assess a correct diagnosis detailed pretreatment work-up is needed in order to avoid misdiagnosis and undertreatment. Moreover, complete surgical excision is mandatory, such as a rigorous follow-up. We perform a standard facelift approach with identification and monitoring of the distal facial nerve. There is no evidence of recurrence at 12 months follow-up and no other complications have been noted. Considering that adenoid cystic carcinoma of accessory parotid gland is uncommon, further investigations and ultrastructural studies are needed to establish specific diagnostic criteria.

Ethical approval

Not required.

Declaration of competing interest

The authors have no conflicts of interest to declare.

Acknowledgements

A special thanks goes to Prof. Stefania Staibano and to Dr Silvia Varricchio (Department of Advanced Biomedical Sciences, Pathology Section, University of Naples “Federico II”, Naples, Italy) for the support on the histological description, and for the specimen pictures.

References

[1] Perzik SL, White IL. Surgical management of preauricular tumors of the accessory parotid apparatus. Am Surg 1966;12:498–503.
[2] Frommer J. The human accessory parotid gland: its incidence, nature, and significance. Oral Surg Oral Med Oral Pathol 1977;43.
[3] Toh H, Kodama J, Fukuda J, Rittman B, Mackenzie I. Incidence and histology of human accessory parotid glands. Anat Rec 1993;236(3):586–90.
[4] Barca, I., Novembre, D., Cordaro, R., Lo Faro, C., Colangeli, W., Boschetti, C. E., et al. Myoepithelioma of the parotid gland: a case report with review of the literature. Oral and Maxillofacial Surgery Cases, doi:10.1016/j.omsc.2019.100131.
[5] Luksic I, Mamic M, Suton P. Management of accessory parotid gland tumours: 32-year experience from a single institution and review of the literature. Int J Oral Maxillofac Surg 2019 Sep;48(9):1145–52. https://doi.org/10.1016/j.ijom.2019.02.014. Epub 2019 Mar 9. PubMed PMID: 30862411.
[6] Yang X, Ji T, Wang L, et al. Clinical management of masses arising from the accessory parotid gland. Oral Surg Oral Med Oral Pathol 2011;112:290–7.
[7] Johnson F, Spiro R. Tumors arising in the accessory parotid tissue. Am J Surg 1979;138(5):576–8. 671–6.
[8] Klotz DA, Ju Coniglio. Prudent management of the mid-cheek mass: revisiting the accessory parotid gland tumor. Laryngoscope 2000;110(10 Pt 1):1627–32.

[9] De Riu G, Meloni SM, Massarelli O, Tullio A. Management of midcheek masses and tumors of the accessory parotid gland. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;111(5):e5–11.
[10] Romano A, Lo Faro C, Salzano G, Dell'Aversana Orabona G, Petrocelli M, Corvino R, et al. Synovial chondromatosis of temporomandibular joint spreading into the cranial space. JOMSMP 2018;30:267–71.
[11] Iguchi H, Wada T, Matsushita N, Oishi M, Teranishi Y, Yamane H. Evaluation of usefulness of fine-needle aspiration cytology in the diagnosis of tumours of the accessory parotid gland: a preliminary analysis of a case series in Japan. Acta Otolaryngol 2014;134(7):768–70. https://doi.org/10.3109/00016489.2014.905704.
[12] Baklacı D, Güngör V, Özcan M, Yılmaz YF, Ünal A, Çolak A. Adenoid cystic carcinoma of the accessory parotid gland. Kulak Burun Bogaz Ihtis Derg 2015;25(5):302–5. https://doi.org/10.5606/kbbihtisas.2015.45467.
[13] Das S, Nayak UK, Buggavetti R, Sekhar S. Adenoid cystic carcinoma of accessory parotid gland: a case report. J Oral Maxillofac Surg 2016;74(5):1097. e1–.
[14] Dell' Aversana Orabona G, Abbate V, Piombino P, Iaconetta G, Califano L. Midcheek mass: 10 years of clinical experience. J Cranio-Maxillo-Fac Surg 2014;42(7):e353–8.
[15] Rodino W, Shaha AR. Surgical management of accessory parotid tumors. J Surg Oncol 1993;54(3):153–6.
[16] Eveson JW, Cawson RA. Salivary gland tumours. A review of 2410 cases with particular reference to histological types, site, age and sex distribution. J Pathol 1985 May;146(1):51–8. PubMed PMID: 4009321.
[17] Huang M, Ma D, Sun K, Yu G, Guo C, Gao F. Factors influencing survival rate in adenoid cystic carcinoma of the salivary glands. Int J Oral Maxillofac Surg 1997 Dec;26(6):435–9. PubMed PMID: 9418145.
[18] Batsakis JG, Luna MA, el-Naggar A. Histopathologic grading of salivary gland neoplasms: III. Adenoid cystic carcinomas. Ann Otol Rhinol Laryngol 1990;99:1007.
[19] Zbären P, Nuyens M, Caversaccio M, Greiner R, Stauffer E. Postoperative radiation therapy for T1 and T2 primary parotid carci-noma: is it useful? Otolaryngol Head Neck Surg 2006;135:140.
[20] Barca I, Lo Faro C, Cordaro R, Novembre D, Mignogna C, Cristofaro MG. Intramuscular lipoma involving the masticator space: a case report. JOMSMP; 2019. https://doi.org/10.1016/j.ajoms.2019.02.003.