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Case Report

Mucoepidermoid Carcinoma Arising in the Accessory Parotid Gland: A Case Report

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Abstract: Accessory parotid gland (APG) tumour is a rare pathology located at the mid-cheek or infra-zygomatic region. APG is usually away and separated from the main parotid gland. APG lies on the masseter muscle anteriorly and empties into the Stensen's duct via multiple accessory ducts. We report a case of mid cheek swelling associated with APG. In the literature, the prevalence of accessory parotid gland tumours is around 1% to 7.7%. High suspicion is needed for the diagnosis of accessory parotid gland tumours because of its rarity.

Key words: Accessory parotid gland; Mucoepidermoid carcinoma; salivary gland tumour.

Introduction

The differential diagnosis of swellings in the mid-cheek area include lesions originating from normal anatomical structures as well as from accessory parotid gland (APG) tissue[1]. APG is small salivary gland tissue that is separated from the main body of the parotid gland. It is lying anteriorly on masseter muscle and has its own duct system which is connected to the Stensen's duct[2]. It is therefore very important to differentiate from the anterior facial process of parotid gland, which is part of the main parotid gland but not totally separated from the main gland. In normal anatomical variants, APG is found in around 21% – 69% of individuals[2,3]. It is possible that this anatomical variant will go unnoticed on regular inspection, mainly during its natural glandular state. However, it may undergo same pathological processes as the main parotid gland, resulting in a development of a mid-cheek lump[1,4].

Tumours from APG can be benign or malignant. APG tumours account for only 1% – 7.7% of all parotid gland tumours[1,4]. It was assumed that the behavior and spectrum of these lesions are like tumours that originated in the main parotid gland. In this report we would like to present a case of such a presentation in our center.

Case Report

A 37-year-old patient presented with a slow growing swelling on the right cheek in the past 3 years (Figure 1). 2 months prior to his presentation to us, he developed mild pain at the site of complaint. He was well, able to tolerate orally with no mealtime syndrome. No further symptoms were mentioned. Patient is known to have Type I diabetes mellitus which was diagnosed at age of 11 years old and currently on insulin. He smokes and consumes alcohol occasionally. No history of malignancy in family members was reported. He denies any unintentional loss of weight or poor appetite over the past few years.

On clinical examination, we noticed a swelling at the right mid-cheek region, oval in shape measuring 4 × 3 cm. The swelling is slightly tender, non-mobile and firm on palpation. Surrounding skin appears normal in color. Facial nerve function was found to be normal. Mouth opening is good without any functional disturbances. He has no palpable cervical lymph nodes. Intraoral examination found no obvious swelling, but palpable mass as described was evident on bimanual palpation. Right Stensen's duct opening appears patent without any inflammation of surrounding mucosa. However, slight mucoid secretion was noted compared to saliva secretion on left side.

Ultrasound examination revealed a well-defined predominantly solid heterogeneous hyperechoic lesion in the right cheek. The lesion has foci of calcifications and internal vascularity within. The most anterior aspect of the right parotid gland is abutting the lesion. Ultrasound findings were suggestive of a solid soft tissue lesion with benign features. Magnetic resonance imaging (MRI) scan was later performed and showed a well-defined enhancing solid lesion measuring 35mm × 24mm × 25 mm in the anterior part of right masticatory space overlying anterior part of right masseter muscle and overlying the zygomatic muscle. It extends from anterior margin of right parotid gland to anterior margin of

Figure 1. *Frontal view (A), worm view (B) and lateral view (C) of the right mid-cheek swelling.*

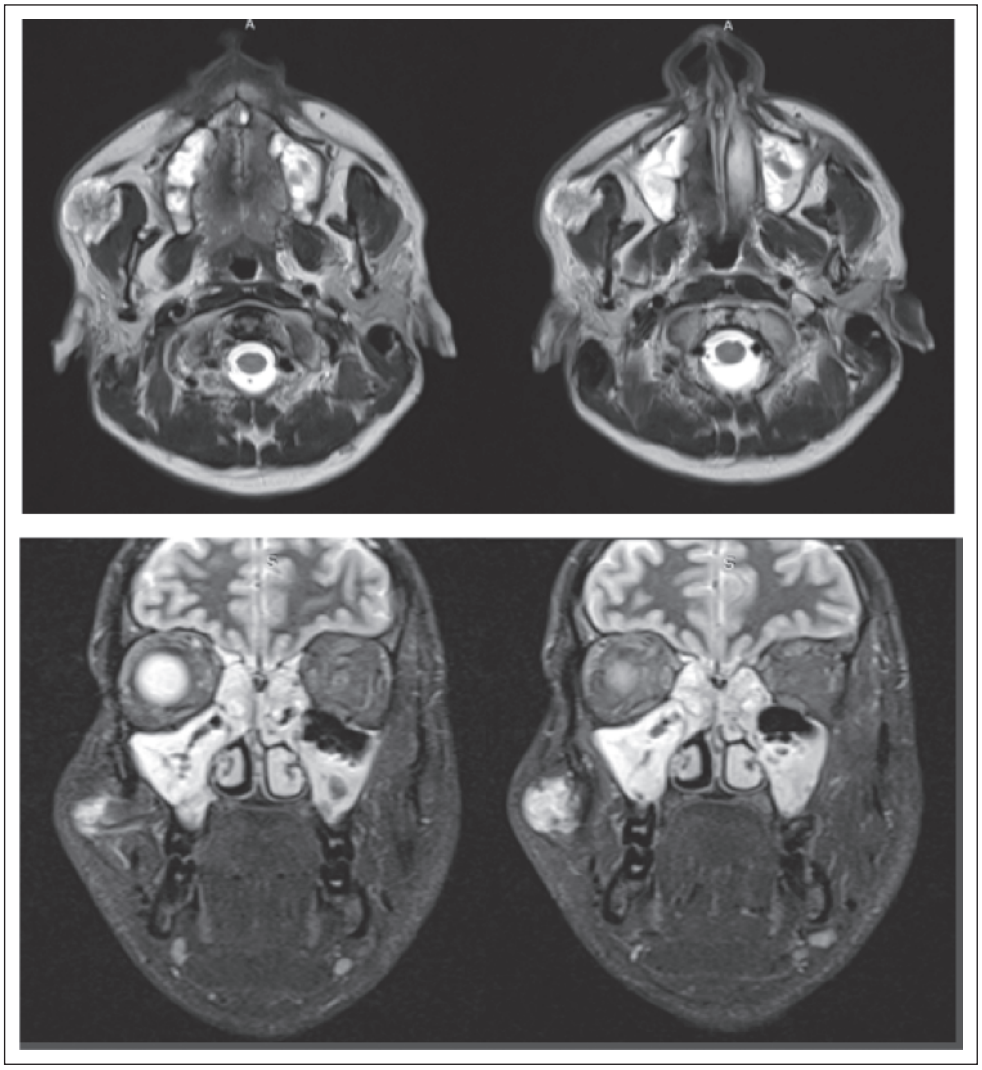

Figure 2. *MRI images from axial view (upper) and coronal view (below). Noted the mass is blended with the right Stensen's duct.*

masseter muscle with slight enhancement of right parotid duct (Figure 2).

Impression from MRI is suggestive of right masseter muscle lesion (myoma/sarcoma), APG lesion (pleomorphic adenoma) or other connective tissue tumour. Therefore, an excisional biopsy under general anesthesia was planned. Excision was done via transoral approach. Intraoperatively, there was no clear demarcation or tissue plane seen between the mass and surrounding masseter muscle. Postoperatively, he developed weakness of the right buccal branch of facial nerve which subsequently resolved after 1 month. Histopathology examination (HPE) reported the lesion as mucoepidermoid carcinoma intermediate grade with positive margin (Figure 3). Due to the findings from the HPE, he was suggested for further surgical treatment with superficial parotidectomy via pre-auricular approach. However, he was very reluctant for another operation. He was also seen by an oncologist and given a second option of having radiotherapy which he refused and was given a monthly follow-up. He subsequently did not turn up for the follow-up.

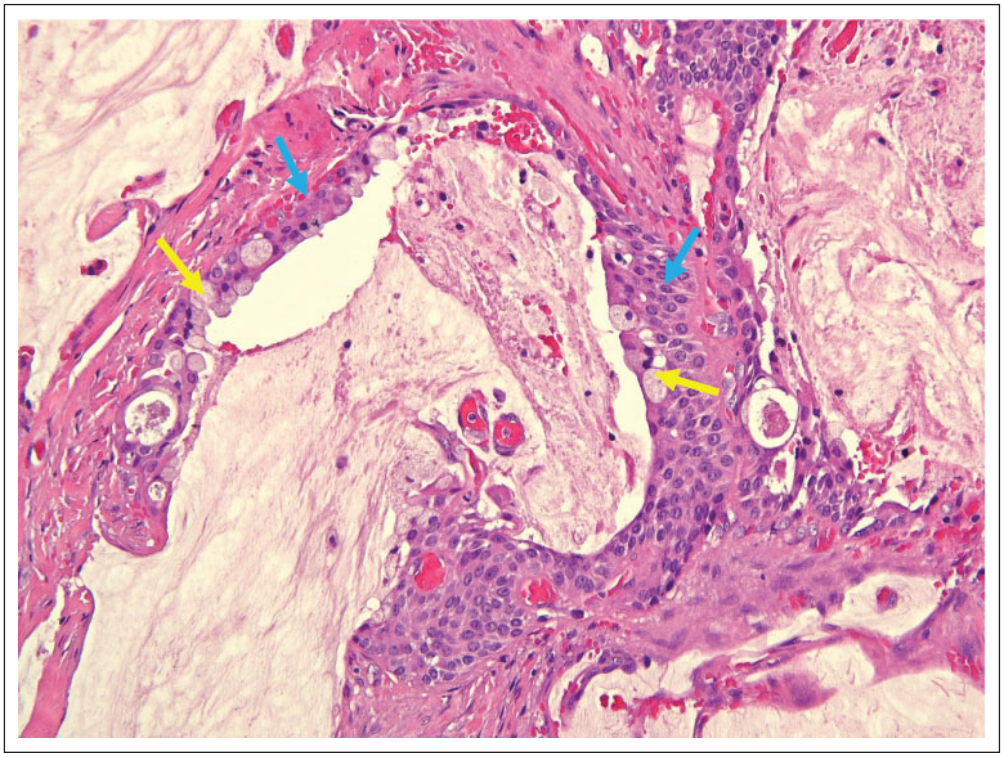

Figure 3. *HPE image at magnification ×200 shows presence of mucous cell (yellow arrow) and epidermoid cells (blue arrow).*

Discussion

The APG is a salivary gland tissue which is separated from the main parotid gland. It is located between the skin and the masseter muscle and approximately 6 mm in front of the main parotid gland, along an imaginary line running from the tragus to the halfway point between the nasal ala and the vermilion border of the lip[5]. It has a secondary duct which empties into the duct of Stensen[3]. The size range of APG is from 0.5–1 cm diameter in size[2].

The prevalence of APG in the normal population is around 21% to 69%[2,3,6,8]. The APG is located between or deep inside the facial nerve's zygomatic and buccal branches. This gland is closely related to the Stensen's duct. The APG can become noticeable as a mid-cheek lump in pathologic states secondary to underlying disease processes, although usually clinically undetectable[6]. The neoplasms found in APG are typically present as slow-growing, painless, mid-cheek swellings. In the mid-cheek area, assessment of a mass may be challenging. In this region, lesions can result from any part of facial soft tissues, including skin, lymph, nerve, vessels, and salivary structures. Skin lesions (epidermoid cyst, sebaceous cyst, fibrosis), arteriovenous malformation or tumours, parotid gland cyst, sialodenitis, duct pathology (duct stone, cyst or tumour), neural lesions (schwannoma), haematoma, lymphadenopathy, benign and malignant salivary gland tumour (salivary carcinoma, lymphoma, sarcoma) and metastasis are differential diagnoses in this area[7].

The reported prevalence of APG tumour is 1% to 7.7% of all parotid gland tumours. APG tumours are uncommon, and benign tumours of the accessory parotid gland account for around 50% to 74% of all APG tumours[5,9]. However, the prevalence of malignancy in APG tumour varies from 26% to 50%[1,4]. The prevalence of malignancy is much lower in the main parotid gland, at approximately around 18.5 percent[3].

A study by Toh et al[3] found that about 27% of APG have mixed acini on histopathological examinations. As comparison to the main parotid gland, the predominant acini are serous acini. Our clinical examinations found that upon milking of the swelling, there is mucoid secretion from Stensen's duct opening which suggests the possibility of APG being composed of mixed acini. Malignant salivary gland tumours are associated with size of the gland; the smaller the gland, the higher the prevalence of malignant tumour[9]. Mixed acini has been thought to be associated with increased malignant tumour occurrence[3]. Due to the sporadic existence of APG in the healthy population, a broad range of lesions need to be considered in the differential diagnosis of an anterior cheek mass. It is found that most of these pathological conditions has normal salivary flow, making clinical diagnosis even more difficult. In our case, mucoid secretion was observed at the right Stensen's duct opening. Thicker saliva flow was seen compared to the left side. No inflammation surrounding the duct's opening was observed. Pathology associated with salivary glands should be suspected and tumour of APG should be included in differential diagnosis in any patient presenting with a mid-cheek mass.

A thorough physical examination, CT scans, MRI, ultrasound scans, and preoperative fine-needle aspiration (FNA) biopsy should be included in the preoperative assessment. All investigations can help to develop the diagnosis. The risk and complications of malignancy due to the relatively very low incidence of lesions in this area are hard to convey to patients. Imaging methods such as MRI, CT and ultrasound help to identify the lesion anatomically, but have very low sensitivity to differentiate between benign and malignant lesions. In the examination of parotid gland tumours with the potential to

Table 1. *Comparison of tumours between main parotid gland and accessory parotid gland*

Comparison	Main parotid salivary gland	Accesory Parotid Gland	Comments
Age	Older patients[14,15] (52–58 years)	Younger patients[8,16] (41–42 years)	APG tumour associated with younger populations.
Malignant tumour	13–18.5%[3,6,17]	25%–55.3%[4,8,7,16-20]	The prevalence of malignant tumour is higher in APG tumours compared to main parotid gland tumours.
Most common malignant tumour	Mucoepidermoid carcinoma[16]	Mucoepidermoid carcinoma[6]	Mucoepidermoid carcinoma is the most common malignant tumour at both sites.
Benign tumour	77%[16]	70%[6]	APGs has lower prevalence for benign tumours compared to parotid gland tumours.
Most common benign tumour	Pleomorphic adenoma[16]	Pleomorphic adenoma[6]	Pleomorphic adenoma is the most common benign tumour at both sites.
Histology features	Predominantly serous acini	27% of APG associated with mixed acini. Almost resembles submandibular gland histology features[3]	This may explain the higher prevalence of malignant tumours in APG compared to main parotid glands.

differentiate between benign or malignant tumours, FNA is shown to be effective in 81% to 98% of cases[10]. The diagnostic precision of FNA decreased to 48% in larger pooled interlaboratory trials, with a sensitivity of 73% and 91% for malignant and benign tumour detection, respectively. However, the quality of FNA depends on many factors, including centers with large pooled pathological evidence, expertise and experience of the assessing pathologist, and the use of adjunct methods such as immunophenotyping or flow cytometry. Therefore, before making a final surgical decision, both clinical and diagnostic tests such as CT, MRI or ultrasound should be considered[6].

Typically, APG tumours occur as mid-cheek masses without symptoms. There is a tendency to excise these masses locally. The risk of injury to branches of the facial nerve is considered to be high and a good exposure for the tumour resection cannot be achieved only by direct transoral approach[8]. However, as suggested by De Riu et al[11], the risk of complications can be minimized even with transoral approach or direct skin incision especially for small or midsized lesions. This is not without contradiction. Toh et al[3] found more than half of the accessory parotid glands have interconnection of the zygomatic and buccal branches of the facial nerve hidden within them, thus this method poses a high risk of nerve injury[3]. For the surgical management of an APG tumour, the standard parotidectomy procedure is very safe, practically easy to perform, reliable, and aesthetically acceptable. This method allows full surgical exposure of the lesion and allows the facial nerve of the distal branches to be identified for nerve function preservation[1]. There are possible risks, including salivary fistula, local recurrence, tumour spillage, sialocele, facial nerve paralysis, and psychosocial impact post-operatively, simply by directly excising APG lesions and primary closure[1,13].

The differential diagnosis of a mid-cheek mass should include APG tumours even if it is rare. Patients with APG tumours were younger than those reported by other authors[7,16]. To avoid misdiagnosis and under-treatment, thorough diagnostic and pretreatment work-up are required. Table 1 shows the comparison between epidemiological data on tumours between main salivary gland and APG.

Conclusion

Compared with its primary parotid gland, APG has a higher prevalence of malignant tumours. Histologically, the characteristics of APG and the main parotid gland can be different. All mid-cheek facial swellings must include accessory parotid gland tumour as one of its differential diagnosis. Thorough clinical examinations, FNAC, ultrasound and MRI or CT scans provide pre-operative evaluation for mid cheek swelling. Approaches to standard parotidectomy are recommended for surgical treatment of the APG tumour, with minimally invasive procedures (direct transoral approach) reserved for benign subtypes.

Declaration

I certify that this case report is original, that it was written by me, and that it has never been published before. The patient's written permission was obtained for the use of clinical images in this case report. During the writing of this case report, no funds were needed.

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References

1. Johnson FE, Spiro RH. Tumours arising in accessory parotid tissue. Am J Surg. 1979;138(4):576-8.
2. Frommer J. The human accessory parotid gland: Its incidence, nature, and significance. Oral Surgery, Oral Med Oral Pathol. 1977;43(5):671-6.
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. Perzik SL, White IL. Surgical management of preauricular tumours of the accessory parotid apparatus. Am J Surg. 1966;112(4):498-503.
5. Lewkowicz A, Levy Y, Zeltser R, Zagury A, Nahlieli O. Accessory parotid gland masses. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2000;89(5):610-2.
6. Lin DT, Coppit GL, Burkey BB, Netterville JL. Tumours of the accessory lobe of the parotid gland: A 10-year experience. Laryngoscope. 2004;114(9 I):1652-5.
7. Newberry TR, Kaufmann CR, Miller FR. Review of accessory parotid gland tumours: Pathologic incidence and surgical management. Am J Otolaryngol – Head Neck Med Surg. 2014;35(1):48-52.
8. Luksic I, Mamic M, Suton P. Management of accessory parotid gland tumours/ : 32-year experience from a single institution and review of the Int J Oral Maxillofac Surg. 2019;48(9):1145-52.
9. Sun G, Hu Q, Tang E, Yang X, Huang X. Diagnosis and Treatment of Accessory Parotid-Gland Tumours. J Oral Maxillofac Surg. 2009;67(7):1520-3.
10. Luk I. Salivary gland tumours/ : 25 years of experience from a single institution in Croatia. J Cranio-Maxillo-Facial Surg. 2012;40:e75-81.
11. Seethala RR, Livolsi VA, Baloch ZW. relative accuracy of fine-needle aspiration and frozen section in the diagnosis of lesions of the parotid gland. Wiley Intersci. 2005; (March):217-23.
12. De Riu G, Meloni SM, Massarelli O, Tullio A. Management of midcheek masses and tumours of the accessory parotid gland. Oral Surgery, Oral Med Oral Pathol Oral Radiol Endodontology. 2011;111(5):e5-11.
13. Klotz DA, Coniglio JU, Hypothesis O. Prudent Management of the Mid-Cheek Mass/ : Revisiting the Accessory Parotid Gland Tumour. Laryngoscope. 2000; (October):1627-32.
14. Dell G, Orabona A. Surgical Management of Benign Tumours of the Parotid Gland/ : Extracapsular Dissection Versus Superficial Parotidectomy – Our Experience in 232 Cases. J Oral Maxillofac Surg. 2013;71(2):410-3.
15. Yu G, Ma D, Zhang Y, Peng X, Cai Z, Gao Y, et al. Multiple primary tumours of the parotid gland. Int J Oral Maxillofac Surg. 2004;33(33):531-4.
16. Ma H, Jin S, Du Z, Wang L, Zhang Z. Pathology and management of masses in the accessory parotid gland region/ : 24-year experience at a single institution. J Cranio-Maxillofacial Surg. 2017;1-7.
17. Eneroth CM. Salivary gland tumours in the parotid gland, submandibular gland and the palate region. Cancer. 1971;27:1415-8.
18. Yang X, Ji T, Wang LZ, Yang WJ, Hu YJ, Zhong LP, et al. Clinical management of masses arising from the accessory parotid gland. Oral Surgery, Oral Med Oral Pathol Oral Radiol Endodontology. 2011;112(3):290-7.
19. Lukšić I, Suton P, Rogić M, Dokuzović S. Accessory parotid gland tumours: 24 years of clinical experience. Int J Oral Maxillofac Surg. 2012;41(12):1453-7.
20. Nemecek JR, Marzek PA, VL Y. Diagnosis and Treatment of Accessory Parotid Gland Masses. Annals of Plastic Surgery. 1994. Jul;33(1):75-79.

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