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Systematic Review

A story of the missing parotid: case report and systematic review of literature

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ABSTRACT

Anatomic variations of the parotid gland are rare. We present the case of a 69-year-old male who presented with unilateral slow growing left sided cheek swelling. Radiological imaging revealed buccal space masses with characteristics similar to those of a salivary gland. Of particular note was the radiological absence of the left parotid gland. Due to the diagnostic uncertainty of this unusual presentation, the patient underwent surgical excision. An intraoral approach was utilized, with careful consideration of the facial nerve's course. Istopathological examination confirmed the diagnosis of a pleomorphic adenoma (PA) of ectopic parotid tissue. A thorough review of literature in PubMed and Google revealed only four previously reported cases, prompting us to include a brief systematic review. Given the rarity of ectopic parotid cases, it is often a diagnosis of exclusion. However, the occurrence of PA within ectopic parotid tissue underscores its importance in the differential diagnosis of unilateral cheek swelling.

Keywords: Ectopic parotid, Parotid agenesis, Pleomorphic adenoma

INTRODUCTION

The parotid gland, the largest of the three paired salivary glands, is typically located in the retromandibular fossa, bordered superiorly by the zygomatic arch, anteriorly by the masseter muscle, and posteriorly by the sternocleidomastoid muscle.[1] These glands develop from ectodermal buds that first appear around the 6th week of intrauterine life near the inner cheek, close to the angle of the mouth, before migrating towards the ear.[2]

Accessory parotid glands, well documented in the literature, are characterized by parotid tissue collections separate from the main gland.[3] These accessory glands are typically located anterior to the main gland, along the course of the buccal branch of the facial nerve, cephalad to Stensen's duct, and superficial to the masseter muscle. While accessory parotid glands are relatively common, congenital agenesis or ectopia of major salivary glands remains a rare occurrence. Agenesis is generally unilateral, with bilateral agenesis being extremely uncommon.[2,4] In cases of agenesis, the absence of the salivary gland may be asymptomatic or lead to symptoms such as xerostomia, halitosis, and oral candidiasis.

Ectopia of the salivary system refers to the presence of salivary gland tissue-whether as a complete organ or disorganized tissue-located outside its normal anatomical position.[5] Although there are some case reports that document the presence of ectopic parotid tissue, fewer describe the association of PAs with ectopic parotid glands. In this report, we present a case of PA within an ectopic parotid gland and offer a review of the existing literature on the development of PAs in ectopic parotid tissue.

METHODS

The PRISMA (Preferred reporting items for systematic reviews and meta-analyses) guidelines were followed. The literature search was based on all case reports or literature reviews involving presence of PA in ectopic

parotid tissue published till date found using PubMed and Google search engine.

Inclusion criteria

Cases with unilateral or bilateral ectopic parotid gland, presence of PA within the ectopic parotid gland and case reports, literature reviews, case series were included.

Exclusion criteria

Cases with heterotopia and cases with ectopic parotid without the presence of PA were excluded.

A 69-year-old male presented to department of ENT, GMCH Nagpur, with recurrent gradually increasing painless left cheek swelling for 8 years. He had history of excision of similar swelling intraorally 10 years ago. On inspection there was a single swelling 3×3 cm size (Figure 1). On palpation two independent swellings were palpated. The larger one, size 3×3 cm lying in the left buccal space and smaller swelling, size 2×2 cm lying anterior to larger swelling. Both swellings were firm in consistency, nontender, freely mobile in all directions within the buccal space.

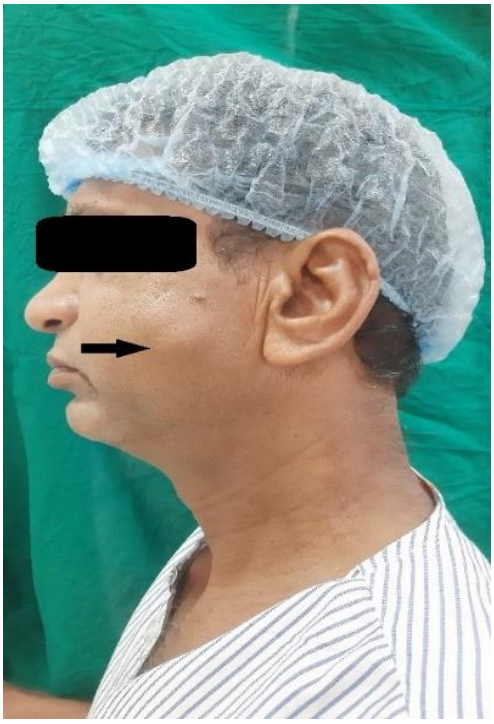

Figure 1: Clinical photo.

MRI (Figure 2) revealed two separate well defined multilobulated lesions in buccal space, hypointense on T1 and iso-hypointense on T2/STIR with heterogenous contrast enhancement probably ectopic of parotid. Normal left parotid gland was absent. Its bed was replaced by fatty tissue. FNAC findings from both the swelling was PA. A diagnosis of PA of ectopic of parotid with unilateral agenesis of normal gland was made.

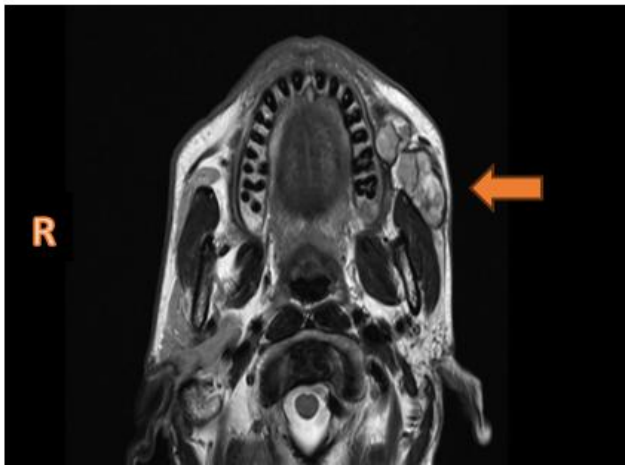

Figure 2: Axial MRI showing tumor.

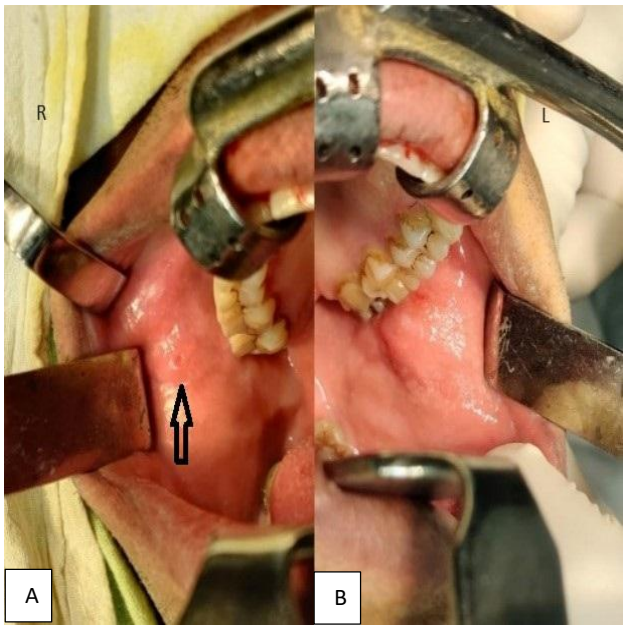

Figure 3 (A and B): Stensen's duct papilla absent on side of tumor.

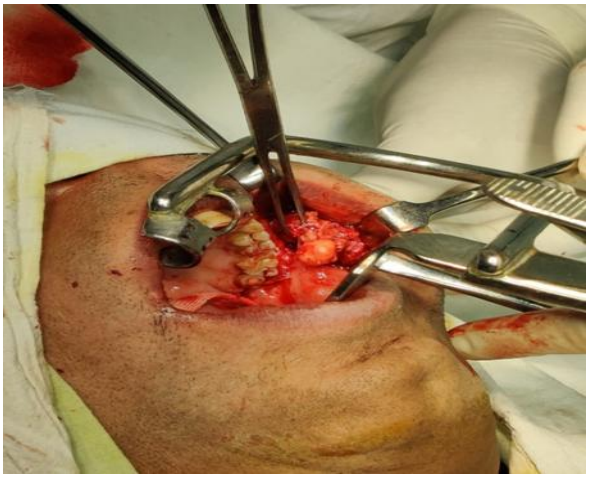

Figure 4: Intraoperative photo of tumor.

Patient was then subjected to intraoral extracapsular excision of tumor. Intraoperatively the absence of Stensen's duct papilla was noted on the abnormal side (Figure 3 and 4). The post operative histopathology report confirmed PA.

RESULTS

A total of four case reports meeting the inclusion criteria were identified, bringing the overall number of documented cases to five (n=5), including our own case. To date, there have been no literature reviews on this topic, making our report likely the first of its kind. Almost all cases (4/5) presented with a slow-growing, asymptomatic cheek mass, with the exception of one case, where the presentation involved an anterior neck mass. In four cases, the ectopic parotid tissue was found ipsilateral to the absent normal tissue, while in one case, it was contralateral. All cases underwent radiological examination and fine needle aspiration cytology (FNAC). The most commonly used imaging modality was MRI, employed in all 5 cases, followed by CT in 3 cases. Tc99 scintigraphy was used in two cases to detect functioning salivary glands. All patients underwent surgery, and postoperative histology revealed benign PAs in all cases. These results are summarized in Table 1 for convenience.

Table 1: Literature review of association of PA with ectopic parotid gland.

Authors (year published)	Presentation	Status of parotid gland	Radiology undertaken	Treatment	Histology
Cotelingam et al[6] (1983)	Anterior mid-cervical mass			Surgery	PA of ectopic parotid
Karakoc et al[7] (2005)	Slowly enlarging left parotid mass	Absent right parotid gland	CT, MRI, Tc99 scintigraphy	Subtotal parotidectomy	PA
Lee et al[8] (2010)	Slowly enlarging right cheek mass	Absent right parotid gland	CT, MRI	Surgery	PA
Seith et al[9] (2013)	Painless left cheek swelling	Absent left parotid gland	CT, MRI, Tc99 scintigraphy	Surgery	PA
Our case	Painless left cheek swelling	Absent left parotid gland	MRI	Lumpectomy	PA

DISCUSSION

The true incidence of agenesis or ectopia of the parotid gland remains difficult to determine, as these conditions are often asymptomatic. Congenital agenesis of the parotid gland is frequently associated with other anomalies, including branchial arch abnormalities and craniofacial deformities.[10] Bilateral agenesis of the parotid gland has been reported in cases of Lacrimoauriculodentodigital (LADD) syndrome, a condition linked to autosomal dominant inheritance.[11] In contrast, unilateral agenesis is rare. A literature review by Teymoortash et al identified 21 cases of unilateral agenesis.[12] They themselves witnessed a case in which a patient presented with a lump over the right parotid area, which, upon investigation, was found to be a lipoma replacing the normal right parotid gland. Gulati et al conducted a literature review of incidental findings of absent or ectopic parotid glands, reporting 22 cases, many of which presented with unilateral, painless swellings of various pathologies, including angioma, branchial cleft cysts, and sialosis.[13] Among these, three cases of PA were noted-one in the contralateral parotid gland and two in ipsilateral ectopic parotid tissue, which are included in our review.

A key observation in many of the reported cases, as reviewed by Teymoortash et al is the absence of the duct papilla in ectopic parotid tissue. Goldenburg et al emphasized that the absence of the duct papilla can be a clinical indicator suggesting the potential absence of the gland itself.[2] Our case also demonstrated the absence of the papilla on the side of the lesion, reinforcing this as a potential diagnostic sign of agenesis with ectopia.

While ultrasound may serve as a primary imaging modality, the importance of cross-sectional imaging, particularly MRI and CT scans, cannot be overstated, especially if surgical management is considered. These imaging techniques provide valuable information regarding the presence or absence of normal parotid tissue, its relationship to the facial nerve, and the presence of other salivary glands. Tc99 scintigraphy can also help identify heterotopia in cases of ectopic tissue. However, many reports in the literature, have not provided details on the relationship between ectopic parotid tissue and the facial nerve, which is crucial for surgical planning. In our case, we approached the tumor intraorally and did not encounter any branches of the facial nerve. Postoperatively, the patient experienced no immediate complications, and follow-up for one year revealed no long-term issues or recurrence. This lack of intra-operative findings, long-term follow-up and recurrence data is a limitation in many studies.

Histologically, the parotid gland typically contains serous acini. However, a cadaveric study on ectopic parotid glands revealed the presence of mixed acini, containing both serous and mucinous elements.[14] This mixture of acini, persisting from early stages of development, may influence the pattern of tumor development within

ectopic parotid tissue. In our case, histopathological examination revealed a PA with normal salivary gland tissue at one end. To date, there has been no documentation regarding the association of malignant tumors with ectopic parotid tissue, which remains an important area for further investigation.

CONCLUSION

Our study contributes to the limited number of cases documenting the occurrence of PA in the presence of ectopic parotid tissue, as reported in the literature. Although rare, the occurrence of PA within ectopic parotid tissue should be considered a key differential diagnosis in cases of cheek swelling. Further research is needed to better understand the nature of tumors associated with ectopic parotid tissue, their impact on the anatomy of the facial nerve and other salivary glands, and their long-term behavior.

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