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CASE REPORT
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ECTOPIC PLEOMORPHIC ADENOMA OF BUCCAL SPACE: CASE REPORT WITH REVIEW OF LITERATURE

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ABSTRACT

Introduction: Salivary gland neoplasms are rare, accounting for 3% to 5% of all head and neck tumours. Pleomorphic adenoma (PA) is the most common benign neoplasm, mainly affecting the major salivary glands; however, its origin from minor salivary glands such as cheek mucosa is sporadic and rarely encountered. Through this clinical report, we share and discuss our experience about this rare entity and highlight the cases, which have been reported in literature until date.

CASE HISTORY

A 47-year-old male labourer by occupation, presented with swelling over the right cheek for 6 months. The swelling was insidious in onset, gradually progressive, initially the size of a pea and had grown to the current size at presentation. On palpation, the swelling was solitary, hard, non-tender, non-mobile, and well localized with smooth margins having no induration around. To confirm the clinical findings a contrast-enhanced CT scan was performed which showed a heterogeneously enhancing 4*4 cms solid, nodular lesion involving the right buccal space. An excision of the lesion was done using an intraoral approach and the histopathology revealed it as pleomorphic adenoma of minor salivary gland.

Conclusion: Although the occurrence of PA in the minor salivary glands of the cheek mucosa is uncommon, the early establishment of a correct diagnosis and initiation of appropriate treatment are important because they permit effective management of the condition and improvement in the prognosis of patients.

KEYWORDS

Pleomorphic adenoma, minor salivary gland, cheek swelling, buccal mucosa

INTRODUCTION

The head and neck region is divided into numerous small tissue spaces by the condensation of deep cervical fascial layer. The infrastructure of the cheek is formed by the buccal space, a tiny potential space on the lateral part of the face 1.

The buccal space anatomically is a small 3 dimensional tissue space bounded by the angle of mouth anteriorly, the masseter muscle posteriorly, zygomatic process of the maxilla and zygomaticus muscles superiorly, depressor anguli oris muscle and the deep fascia attaching to the mandible inferiorly, buccinator muscle

medially and platysma with subcutaneous tissue laterally 3. Buccal space contains various important structures such as adipose tissue, parotid duct, facial artery and vein, small minor salivary glands, lymphatic channels, and branches of the mandibular and facial nerves 4. Swellings in the buccal space are rare to encounter and due to their diverse presentations, they often pose difficulty for the treating surgeons and pathologists to plan their effective management.

Approximately, 8% of pleomorphic adenoma (PA) involves the minor salivary glands and the hard palate is found to be the most common site of origin (60–65%) 5. PAs are known to occur in other minor salivary gland sites, including the lip, buccal mucosa and tongue 6. However, PA of a buccal minor salivary gland, which lies on the external aspect of buccinators, has rarely been reported until date. Clinically, patients often present with a painless, palpable mass in the cheek that may gradually increase in size over time.

A thorough review of existing literature has revealed only a few well-documented cases of PA involving the buccal minor salivary gland. Through this clinical report, we share and discuss our experience about this rare entity and highlight the cases, which have been reported in literature until date.

CASE REPORT

A 47-year-old male patient labourer by occupation, presented at our tertiary care centre with complaints of swelling over right side of the cheek since 6 months. The swelling was insidious in onset, gradually progressive, initially the size of a pea and had grown to the current size of approximately four cms. Patient had no significant medical comorbidities or addiction history. On extra oral inspection a solitary dome-shaped, oval swelling with a smooth surface was present over the right cheek region. The swelling measured approximately 4 × 4 cm in size extending up till the lower border of zygomatic arch superiorly, to the upper border of the mandible inferiorly, posteriorly extending up till the anterior border of masseter and anteriorly 1 cm short of the angle of mouth (Figure 1 A). On palpation, the skin above the swelling was pinchable, and the swelling was firm in consistency, mobile, tender with smooth well-defined margins, non-fluctuant, non-reducible and non-pulsatile (Figure 1 B). There was no associated regional lymphadenopathy.

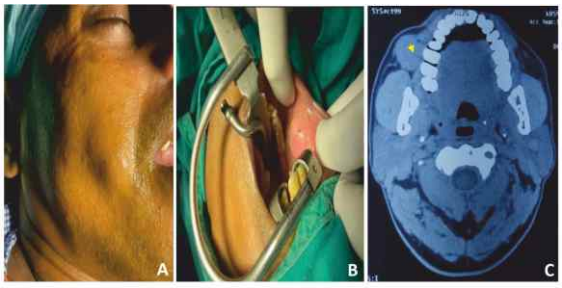

FIGURE 1: A - GLOBULAR SWELLING OVER RIGHT CHEEK

FIGURE 1: B - INTRAORAL EXAMINATION SHOWS 4*3 CMS FIRM, GLOBULAR, TENDER, NON-REDUCIBLE, NON-PULSATILE MASS IN RIGHT BUCCAL MUCOSA.

FIGURE 1: C – CECT REVEALS A SOLID, NODULAR LESION INVOLVING RIGHT BUCCAL SPACE (ARROW)

To further, evaluate the swelling a contrast enhanced computed tomography (CECT) face (figure 1 C) was carried out which was suggestive of a solitary well defined heterogeneously enhancing lesion 3*2 cms in size involving the right buccal space without invasion of the adjacent structures. Fine-needle aspiration cytology (FNAC) from the lesion was carried out to confirm the diagnosis, which revealed chondromyxoid stroma with spindle cells observed embedded in a stromal matrix, and cells were seen in cohesive clusters suggestive of pleomorphic adenoma.

After confirmation of diagnosis, the patient underwent excision of the lesion by intraoral approach under general anaesthesia. After achieving adequate cheek retraction using a mouth gag, a vertical incision intraorally was given over the mucosa covering the swelling in the right buccal space (figure 2 A). Careful dissection was done all around the lesion and with gentle traction; the lesion was excised out en-bloc and sent for histopathological examination (figure 2 B). The buccal space was carefully examined for any remnant and after achieving haemostasis, the defect was closed in two layers. Histopathological examination revealed epithelial , myoepithelial and stromal component with epithelial cells arranged in cords, sheets, nests and islands suggestive of pleomorphic adenoma of minor salivary glands (figure 3A,B). Post-operative period was uneventful and no signs of recurrence until date.

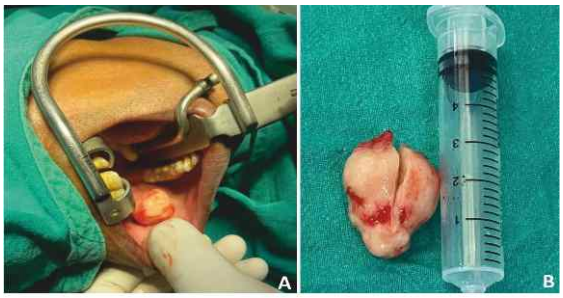

FIGURE 2: A - INTRAOPERATIVE EXPOSURE VIA INTRAORAL APPROACH SHOWING A WELL CIRCUMSCRIBED GLOBULAR MASS IN RIGHT BUCCAL MUCOSA

FIGURE 2: B - SPECIMEN SHOWING GREY WHITE TUMOR MEASURING 3X2CMS. CUT SECTION SHOW SOLID GREY WHITE LESION WITH FOCAL CYSTIC AND HEMORRHAGIC AREAS.

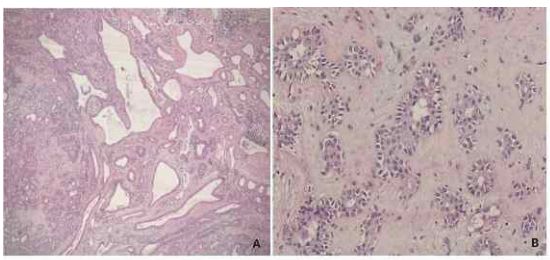

FIGURE 3 A : EPITHELIAL , MYOEPITHELIAL AND STROMAL COMPONENT WITH EPITHELIAL CELLS ARRANGED IN CORDS, SHEETS, NESTS AND ISLANDS (10 X).

FIGURE 3 B : MYOEPITHELIAL CELLS SEEN INTERSPERSED IN FIBROMYXOID STROMA (40 X).

DISCUSSION

Tumours of the salivary glands represent less than 5% of all head and neck tumours and two-thirds of these tumours are pleomorphic adenomas. PA is rarely encountered involving minor salivary glands and occurs approximately in 8% of the cases 6.

The ectopic sites reported for the occurrence of PA include lip, buccal mucosa, floor of mouth, palate and tongue 7. There are 800–1,000 minor salivary glands located throughout the oral cavity in the tissue of the buccal, labial and lingual mucosa; the soft palate; the lateral parts of the hard palate; and the floor of the mouth. Unlike the major glands, they are not encapsulated by connective tissue, but present as clusters of several acini connected in a tiny lobule 8. The glandular lobules are 1–5 mm in diameter and are separated by thin connective tissue. A minor salivary gland may have a common excretory duct with another gland, or may have its own excretory duct. Their secretion is mainly mucous (except for Von Ebner glands) 9.

Patients diagnosed with benign minor salivary gland pathologies usually present with complaints of a small, painless, quiescent nodule, which slowly begins to increase in size, sometimes showing intermittent growth. The skin rarely ulcerates even though these tumours may reach a very large size. Unlike pain being a presenting complaint in our reported case, these swellings are commonly encountered as painless bumps, with associated local discomfort during mastication and speaking 10.

Grossly, they are usually encapsulated, solitary, well-defined, ovoid or round masses. Larger neoplasms may have a characteristic bosselated surface with necrotic or cystic regions. Their consistency varies from hard to rubbery to soft swelling that may be fluctuant. The cut surface of the tumour is characteristically solid and the colour varies from grey blue, pale yellow to tan. There may be gritty areas and gelatinous or glistening foci may be present when there is cartilaginous or myxochondroid differentiation 11.

Lesions arising in the buccal space may be de novo or may be due to the spread of lesions from the adjacent sites. The differential diagnosis of PA cheek includes buccal abscess, dermoid cyst, sebaceous cyst, neurofibromas, lipoma, mucoepidermoid carcinoma and polymorphous low-grade adenocarcinoma 12.

The differentials can be ruled out by thorough clinical, radiological and pathological examination. The buccal space abscess present with signs of inflammation, which can be ruled out as they were absent in the present case. The solid nature of PA and lack of tissue showing the three germ layers rule out the possibility of a mature dermoid cyst. Sebaceous cyst present as fluctuant masses with an external punctum, which differentiate it from PA. As on histological picture, both epithelial and myoepithelial cells were seen; which ruled out mucoepidermoid carcinoma. The negative slip test clinically and the absence of lipomatous component histologically rule out the possibility of lipoma. The absence of perineural invasion and mitotic figures obscure the chances of polymorphic low-grade adenocarcinoma.

We did a literature search of case reports on pleomorphic adenoma involving buccal space using the keywords: "Pleomorphic adenoma,

AUTHOR + REFERENCE	AGE/ GENDER	SYMPTOMS	SIGNS	SURGICAL APPROACH	HPE	FOLLOW UP & RECURRENCE
Bahbahs et al [13]	22 Y / M	Painless swelling in right cheek since 2 years	1*1 cms firm, globular, non-tender, non-reducible, non-pulsatile mass in right buccal mucosa.	Intraoral excision	Pleomorphic adenoma	Not mentioned
Taufik et al [14]	17 Y / F	Painless swelling in right cheek since 4 years	2 cms firm, globular, non-tender, mobile mass in right buccal mucosa.	Intraoral excision	Pleomorphic adenoma	3 yrs. No sign of recurrence
Sharma et al [15]	40 Y / M	Painless swelling in left cheek since 20 years	4*4 cms firm, lobulated, non-tender, mobile mass in left buccal mucosa.	Intraoral excision	Pleomorphic adenoma	Not mentioned
Afonso RAS et al [16]	65 Y / M	Painless swelling in left cheek since 2 years	2.1*1.3*1.4 cms mobile, firm, solitary mass in left buccal mucosa	Intraoral excision	Pleomorphic adenoma	1 yr. No sign of recurrence
Yong hun kim et al [17]	43 Y / F	Painless swelling in right cheek since 3 months	1*1 cms mobile, firm, solitary mass in right buccal mucosa	Intraoral excision	Pleomorphic adenoma	Not mentioned
Periasamy S et al [18]	31 Y / F	Painless swelling in right cheek since 4 years	5*3 cms sessile, firm, mobile, non-tender, non-fluctuant, non-reducible, non-pulsatile swelling in right cheek	Intraoral excision	Pleomorphic adenoma	1 yr. No sign of recurrence
AK Pillai et al [19]	16 Y / F	Painless swelling in left cheek since 4 years	3*2.5 cms mobile, firm, solitary mass in left cheek	Intraoral excision	Pleomorphic adenoma	1 yr. No sign of recurrence

Kalenahalli jagadishkumar et al [20]	9 Y / F	Painless swelling in right cheek since 4 months	2*2 cms mobile, firm, solitary, non-tender, non-fluctuant mass in left cheek	Intraoral excision	Pleomorphic adenoma	1 yrs. No sign of recurrence
Kaul. D et al [21]	36 Y / F	Painless swelling in right cheek since 5 years	1*2 cms well circumscribed, mobile, firm, solitary mass in right buccal mucosa	Intraoral excision	Pleomorphic adenoma	1 yr. No sign of recurrence
Federica Veneri et al [22]	70 Y / F	Painless swelling in right cheek since 15 years	5 cms , firm, mobile, non-tender, hard elastic swelling in right cheek	Intraoral excision with Nd:Yag laser	Pleomorphic adenoma	2 yrs. No sign of recurrence
Andrea avagnina et al [23]	56 Y / M	Painless swelling in right cheek since 15 years	3 cms mobile, firm, well circumscribed, non-tender, non-fluctuant mass in left cheek	Intraoral excision	Pleomorphic adenoma	1 yr. No sign of recurrence
ECF Junior et al [24]	50 Y / F	Painless swelling in left cheek since 4 years	0.5 cms mobile, fibrous, endophytic nodule in the left cheek	Intraoral excision	Pleomorphic adenoma	1 yrs. No sign of recurrence
Idris Cildir et al [25]	80 Y / M	Painless swelling in left cheek since 5 years	4.6*3.1 cms mobile, firm, non-reducible mass in left cheek	Extraoral excision	Pleomorphic adenoma	2 yrs. No sign of recurrence

TABLE 1: LITERATURE REVIEW SHOWING SYMPTOMS, SIGNS, SURGICAL APPROACH AND OUTCOMES OF PLEOMORPHIC ADENOMA INVOLVING BUCCAL MUCOSA

In the case reports reviewed, we observed that the most common symptom reported was painless swelling over cheek ranging in size from 1 cms to 6 cms respectively. As per literature, such lesions have been reported predominantly in females affecting any group, most commonly seen post adolescence. All the reported cases were diagnosed based on preoperative clinical examination, fine needle aspiration cytology and CECT to confirm the pathology. Among all the 13 cases reviewed including our case, complete surgical excision of the tumour was done using an intraoral approach except for 1 case where an extraoral approach was performed 25. Post-operative follow up has been mentioned in 10 cases varying from 1 to 4 years, which showed no signs of recurrence.

PA is known to produce recurrence due to either spillage, inadequate removal or enucleation at the time of operation, but is not known to produce distant metastasis. A recurrence rate of 2–44% has been reported in the literature. The ideal treatment of choice for PA is wide local excision with good safety margins and follow-up for at least 3–4 years.

CONCLUSION

Pleomorphic adenoma of the minor salivary gland especially involving the cheek is a relatively rare lesion and poses a challenge for surgeons and pathologists during planning its management. Pleomorphic adenoma though rare, should always be considered as a differential for buccal space lesions. Surgical excision of the tumour with wide margins is the treatment of choice. So early diagnosis, prompt treatment and regular follow up and close monitoring at least for 5 years are necessary to prevent the risk of recurrence and rare chances of malignant transformation.

DECLARATION

Ethics approval and consent to participate: Ethical approval taken from teh Institute.

Availability of data and material: The datasets during and/or analyzed during the current

study are available from the corresponding author upon reasonable request.

Competing interests: The authors declare that they have no competing interests.

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