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Case Report

Dentistry Section

Buccal Cyst with Respiratory Epithelium: A Diagnostic Dilemma: A Case Report

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ABSTRACT

Teratoid, dermoid and epidermoid cysts are rare, developmental, non odontogenic lesions that are derivatives of germinative epithelium. Epidermoid cysts are most frequently reported in the floor of the mouth, with uncommon occurrence in the buccal mucosa. Dermoid cysts in this location have not been documented and the evidence of respiratory epithelium within such cysts and location is exceedingly rare. A 55-year-old female presented with a painless and slowly enlarging swelling in the left buccal mucosa. Imaging suggested a mucous retention cyst because of its cystic appearance and well-defined margins, but after surgical excision, histopathology revealed a cystic wall partly lined by ciliated pseudostratified columnar epithelium of respiratory type. Such lining is extremely rare in oral cysts and has not been reported in the buccal mucosa. The postoperative phase was uneventful and no recurrence was reported during follow-up. This case is noteworthy as respiratory-type epithelial lining has rare occurrence in oral cysts and that too within the buccal mucosa. This case emphasises the importance of histopathology in confirming unusual variants and highlights the limitations of clinic-radiographic diagnosis and adds to the limited literature on oral cysts with respiratory epithelium.

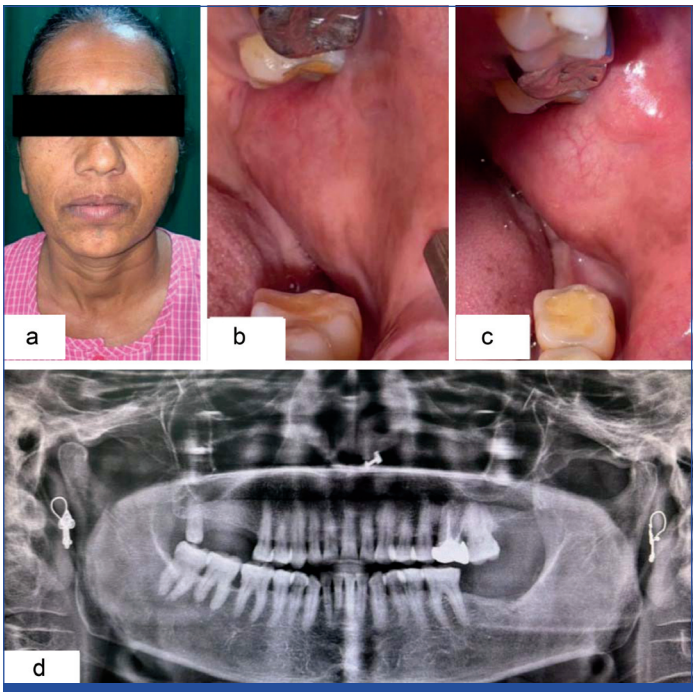
Keywords: Buccal mucosa, Epidermoid cyst, Respiratory epithelium

CASE REPORT

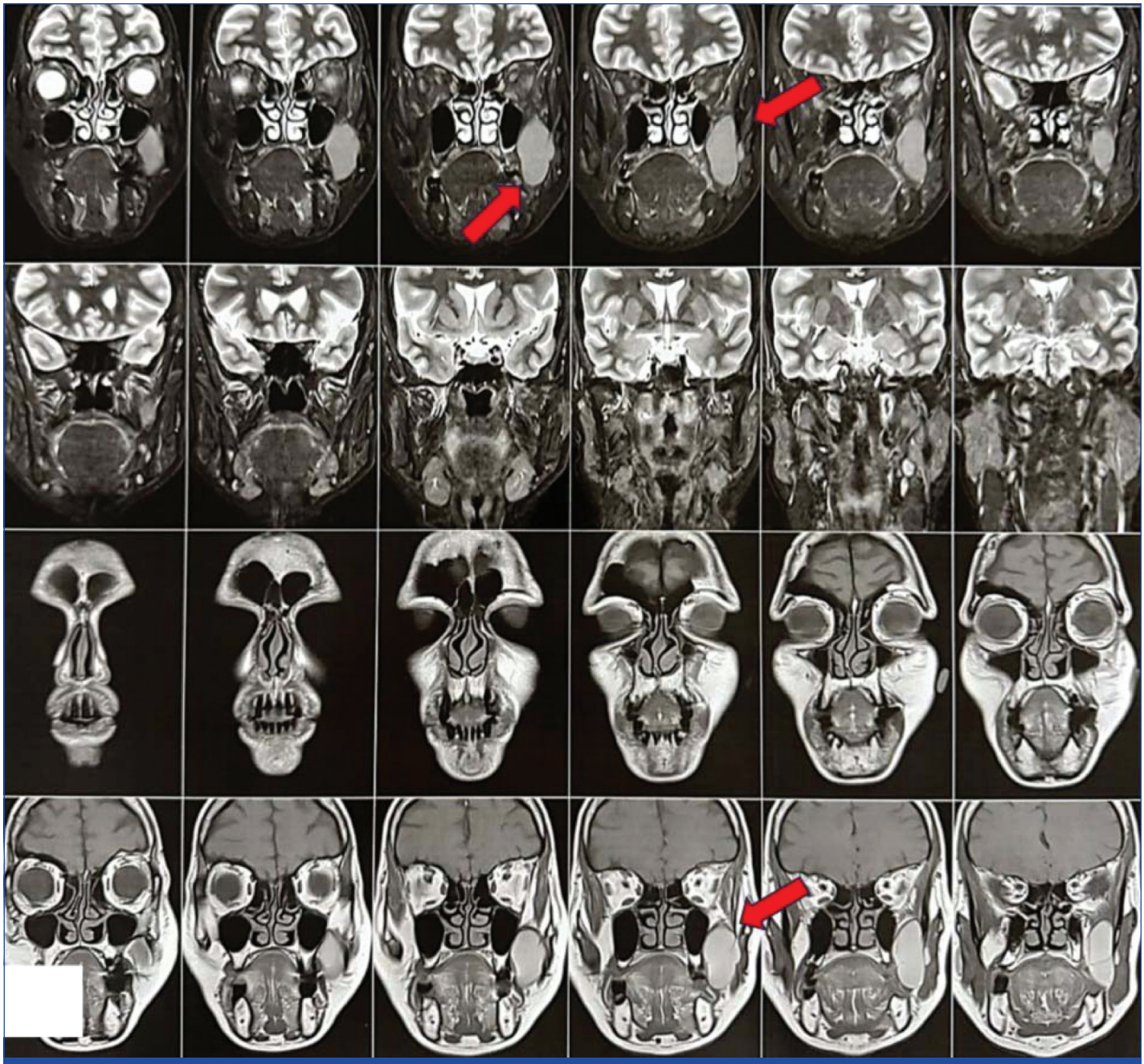
A female, 55-year-old presented to dental Outpatient Department (OPD) with an intraoral asymptomatic swelling on the left cheek for the past two months [Table/Fig-1a]. Patient could not relate the occurrence of swelling to any event such as trauma, during or post-meal or dental complaint. No history of fever or general symptoms were associated. No relevant medical history was associated with the patient. Patient gave history of extraction with #37 and #38 two years back and history of root canal treatment with metal crown prosthesis with #26 five years back. No relevant adverse habit history was noted by patient. No associated extraoral swelling or gross asymmetry was noted.

On examination, an intraoral diffuse, solitary swelling with size of approximately 4×3 cm was noted on left buccal mucosa extending into the posterior region of retromolar area [Table/Fig-1b,c]. On palpation, the swelling was moderately firm in consistency, non tender, non mobile and fluctuant as well as fixed to the skin or underlying tissues. The colour, texture and temperature of the overlying skin were normal. Patient's salivary flow was normal with no signs of increased or decreased salivation. Masseter muscle was normal in palpation. Neck movements were normal. Mouth opening was adequate. Clinical findings were suggestive of provisional diagnosis of epidermoid cyst involving left buccal mucosa. The considered differential diagnoses were benign oral cyst such as mucocele and mature dermoid cyst, benign salivary gland tumour such as pleomorphic adenoma, benign lymphoid proliferation like follicular lymphoid hyperplasia, benign mesenchymal tumours like lipoma, malignant salivary gland tumours such as mucoepidermoid carcinoma.

For further investigations, panoramic radiography was done, that showed no bony involvement [Table/Fig-1d]. Ultrasonography was performed using high frequency linear probe and the findings were suggestive of an anechoic cystic lesion with few incomplete septa of size measuring 39×19×16 mm (Superior-Inferior × Anterior-Posterior × Mediolateral) was noted, indicating mucous retention cyst. For further evaluation, Magnetic Resonance Imaging (MRI) was advised. MRI revealed evidence of 2-3 mm thin-walled lobulated 45×31×28 mm size T2 Weighted (T2W) and T1 Weighted (T1W) bright signal lesion showing bright signals on fat-sat sequence involving left masticator space in close relation with masseter, temporalis and lateral pterygoid muscles on left-side. The lesion appears to be extending to retromaxillary fat pad superiorly and retromolar trigone inferiorly without any stranding or oedema in surroundings. No evidence of internal septations or soft-tissue and no cervical lymphadenopathy were noted on either side. The overall imaging features were suggestive of benign high proteinaceous cystic lesion in masticator space rather than an aggressive or malignant lesion, likely to be mucous retention cyst or minor salivary gland lesion [Table/Fig-2]. Cytology was performed to rule out malignancy, with



[Table/Fig-1]: a) Extraoral photograph; b,c) Intraoral photograph showing solitary swelling in the left buccal mucosa; d) Orthopantomogram showing no evidence of involvement of bone.



[Table/Fig-2]: MRI revealed a well-defined, thin-walled lobulated cystic lesion (45×31×28 mm) in the left masticator space extending into the retromaxillary fat pad and retromolar trigone, without surrounding oedema or cervical lymphadenopathy, suggestive of a benign cystic lesion.

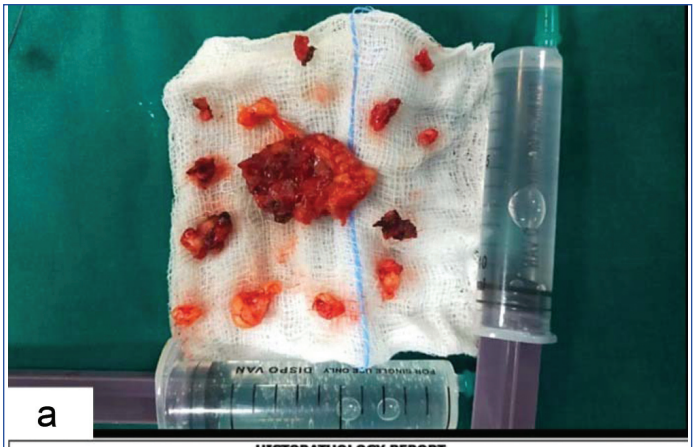
evidence of no malignancy. Complete surgical excision of the lesion was performed with an intraoral approach and was sent to examine histopathologically. On gross examination, the excised specimen was greyish brown cystic measuring 4x3x1.5 cm, external surface was encapsulated and congested [Table/Fig-3a,b].

The Haematoxylin and Eosin (H&E) stained sections with a magnification of 100x of the specimen showed fibro-collagenous cyst wall lined partly by columnar epithelium and partly by ciliated pseudostratified columnar epithelium. Underneath stroma shows lymphocytes, histiocytes and granulation tissue with no nuclear atypia, no dysplasia, no granuloma or malignancy seen [Table/Fig-4a,b]. Histopathological diagnosis of benign oral cyst with respiratory epithelium was achieved. Follow-up was taken after six months and no signs of recurrence were reported. Timeline of history is described in a flowchart [Table/Fig-5].

DISCUSSION

Dermoid, epidermoid, and teratoid cysts are uncommon and occur in the head and neck for 7% and only 1.6% in the oral cavity [1]. The diagnosis of epidermoid cyst poses a considerable challenge for clinicians, as its clinical presentation is often non specific and can closely resemble a variety of other oral pathologies. Consequently, adjunctive investigations such as ultrasonography, fine-needle aspiration, and magnetic resonance imaging are frequently employed to establish a definitive diagnosis and exclude alternative conditions [2]. The occurrence of epidermoid cysts in the buccal mucosa, are extremely rare, and dermoid cysts have not been reported [3]. Teratoid cysts are composed of dermoid elements and also of tissues that are derived from other embryonic origins, such as respiratory and gastrointestinal epithelium, as well as connective components such as striated muscle bundles and distinct deposits of fat [4]. This case report presents an exceptionally atypical existence of cyst occurring in the buccal mucosa with respiratory epithelial lining, this emphasises the importance of diagnosis for such rare entities.

The present case adds a novel dimension to the clinicopathological spectrum by demonstrating respiratory type epithelial lining in a buccal mucosal cyst. Several reports [5-10] have described epidermoid cysts in the buccal mucosa, but none with respiratory epithelium. The presence of respiratory epithelium in oral cysts has been explained by several pathogenetic mechanisms. Congenital misplacement of foregut epithelium during embryogenesis is one possibility, while



a

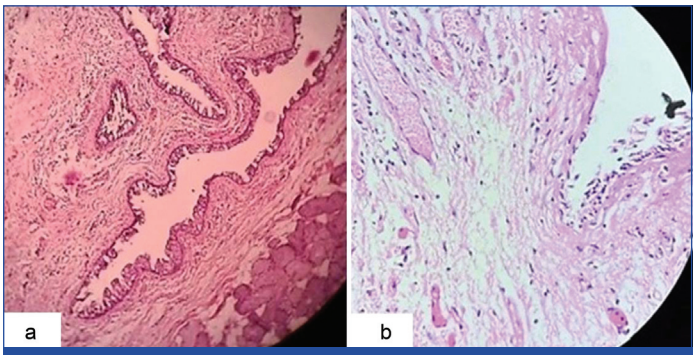
HISTOPATHOLOGY REPORT

HPE no. :	
Clinical details :	
Nature of specimen	**HPE of lesion from left buccal mucosa.**
Gross Examination :	Received specimen labelled as lesion from left buccal mucosa. Received single already cut open greyish brown cystic lesion measuring 4 x 3 x 1.5 cm. External surface - Congested, encapsulated. Cut surface - Grey brown. Sections- 01 and 02 - From cyst (WT).
Microscopy :	H and E stained sections show fibrocollagenous cyst wall lined partly by columnar epithelium and partly by ciliated pseudostratified columnar epithelium. Underneath stroma shows lymphocytes, histiocytes and granulation tissue. No nuclear atypia, no dysplasia, no granuloma or malignancy seen.
Diagnosis :-	**Benign oral cyst with respiratory epithelium.**

b

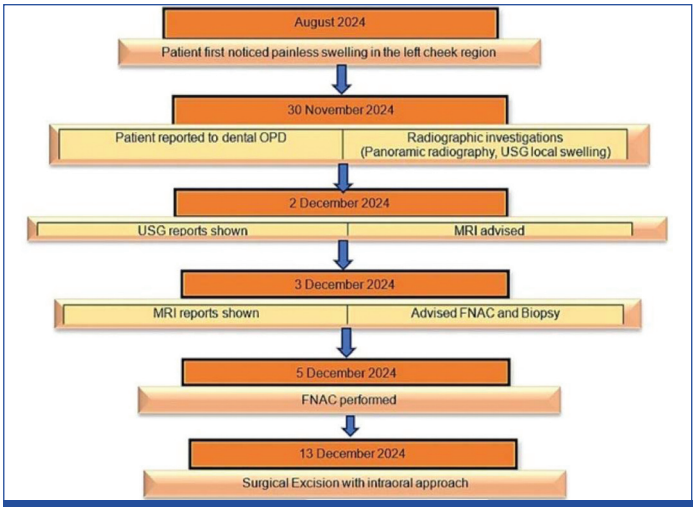
If Specimen preserved, it will be retained in the laboratory for 12 weeks from the date of receipt

[Table/Fig-3]: a) Post-excision specimen; b) Supportive histopathologic report.



[Table/Fig-4a,b]: Fibro-collagenous cyst wall lined partly by columnar epithelium and partly by ciliated pseudostratified columnar epithelium with underneath stroma showing lymphocytes, histiocytes and granulation tissue with no dysplasia, granuloma or malignancy seen (H&E, 100x).

post-traumatic or iatrogenic implantation of respiratory mucosa into oral tissues has been implicated in surgical ciliated Cysts [5]. Respiratory epithelium has been described in other oral Cystic lesions such as lingual cysts with respiratory epithelium (Aforka EE et al., Cialente F et al., [6,8]), teratoid cysts (Palaskar SJ et al., [1]), and



[Table/Fig-5]: Flowchart showing timeline of history.

surgical ciliated cysts of the maxilla (Martinelli-Klay CP et al., Hii EPW et al., [9,10]). These lesions demonstrate ciliated pseudostratified columnar epithelium with goblet cells, sometimes admixed with squamous epithelium [1,6,8-10]. However, none of the published buccal mucosal cysts have shown respiratory lining, making the present case the first documentation of a buccal mucosal cyst with respiratory epithelium. In the current case, the absence of prior maxillary surgery, facial trauma, chronic inflammation, or recurrent infection exempts the possibility of predisposition to epithelial metaplasia. Demonstrating the histopathological examination, a well-differentiated ciliated pseudostratified columnar epithelium was observed, rather than transitional changes that are seen in a metaplastic process. Although, there cannot be complete exclusion of the metaplastic transformation, the absence of known predisposing factors and the histopathological findings favour a congenital development origin in the current case. Clinically, buccal mucosal swellings often mimic mucoceles, salivary gland lesions, or benign neoplasms. Imaging in the present case suggested a mucous retention cyst, consistent with previous report by Ozan F et al., where radiology was non specific [7]. Histopathology therefore remains the gold standard for diagnosis, especially in rare variants. Literature reviews have documented only a handful of epidermoid cysts in this location, and none with predominant respiratory epithelium. Thus, the present case expands the spectrum of oral cystic lesions by demonstrating a benign oral cyst in the buccal mucosa with respiratory epithelial lining. Few studies with oral cysts with respiratory epithelium are reported in the literature and their comparison with the present case is tabulated together [Table/Fig-6] [1,5,6,8-10].

CONCLUSION(S)

The present case presents a rare benign oral cyst of the buccal mucosa that exhibits respiratory epithelial lining, a finding which

Author (Year)	Site	Age/Sex	Histopathological features	Proposed origin	Key findings
Martinelli-Kläy CP et al., [9] (2017)	Maxilla	35 Y/M	Cyst lined exclusively by respiratory epithelium with ciliated and rare mucous cells were also observed.	Surgical ciliated cyst	Respiratory epithelial-lined cyst occurring after maxillary surgery
Hii EPW et al., [10] (2024)	Maxilla	53 Y/F	Multicystic lesion lined by pseudostratified ciliated columnar epithelium.	Surgical ciliated cyst	Respiratory epithelial-lined cyst occurring after orthognathic surgery
Aforka EE et al., [6] (2025)	Tongue	4 Y/M	Respiratory epithelium with goblet cells	Developmental	Lingual cyst with respiratory epithelium; rare congenital lesion
Cialente F et al., [8] (2021)	Tongue	44 Y/M	Cystic lesion lined by well-differentiated ciliated, pseudostratified, columnar epithelium	Developmental	A case with respiratory-type origin of the epithelial cell lining
Palaskar SJ et al., [1] (2014)	Oral cavity (teratoid cyst)	2 Y/F	Respiratory epithelium admixed with other germ-layer derivatives	Developmental/ teratoid	Respiratory epithelium identified within a teratoid cyst
Boffano P et al., [5] (2009)	Oral cavity	35 Y/F	Pseudostratified ciliated columnar epithelium, interspersed by a few goblet cells	Developmental	Surgical management of oral cyst with respiratory epithelium
Present case	Buccal mucosa	55 Y/F	Stratified squamous epithelium with focal ciliated pseudostratified columnar respiratory epithelium	Likely developmental	First reported buccal mucosal cyst showing respiratory epithelial lining

[Table/Fig-6]: Reported oral Cysts with respiratory epithelium in the literature and comparison with the present case [1,5,6,8-10].

has not been previously reported specifically in this location. Its non-specific clinical and radiological presentation underscores the importance of histopathological evaluation for accurate diagnosis. Reporting such unusual variants contributes to a better understanding of the spectrum and pathogenesis of oral cystic lesions.

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