



## Case report

## Non-pancreatic pseudo cyst of oral cavity-case report

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## ABSTRACT

**Introduction and importance:** Cyst is commonly found in oral and maxillofacial region, but non-pancreatic pseudo cyst in this region is quite rare. None of the such cases have been reported so far. Presented here is the case of non-pancreatic pseudo cyst in the right cheek. A description and management of this pathology through open surgery is given, while preserving the anatomy of the cheek.

**Case presentation:** The authors report a non-pancreatic pseudo cyst of the right cheek in a 4 years old boy. His parents noticed swelling over right cheek which slowly increases in size without any other associated symptoms. The swelling was firm, non-tender, nonfluctuant, non-pulsatile, margin was distinct, overlying mucosa was normal in colour, aspiration was negative, 3x2x1 cc in size. The surgical excision of the tumour was performed through an intraoral approach under general anaesthesia. Intraoperatively we found clotted blood confined within fibrous capsule. During one year postoperative follow-up there was no sign of recurrence.

**Clinical discussion:** Pancreatic pseudocysts are benign soft tissue lesion occurring most commonly in pancreas. They are rarely encountered in the soft tissue of Oral and Maxillofacial region. It is the first case of non-pancreatic pseudocyst found in soft tissue of oral and maxillofacial region. The etiopathogenesis of these pseudocysts is not known yet. It may be considered as soft tissue counterpart of Aneurysmal Bone Cyst (ABC).

**Conclusion:** Non pancreatic pseudo cysts may form in soft tissue of oral and maxillofacial region.

## 1. Introduction

Cyst in Oral and Maxillofacial region is common finding. Depending on odontogenesis they are classified into two major types: odontogenic cysts and non-odontogenic cysts [1]. Odontogenic cysts are characterized by specific odontogenic markers, histological similarities with odontogenic structures and anatomical considerations [2]. Non odontogenic cysts originate from specific region of the oral and maxillofacial region like salivary cysts, nasopalatine duct cysts and nasolabial cysts. It also includes some cysts, that may be present anywhere in the body like aneurysmal bone cyst, dermoid cyst and lymphoepithelial cyst [3]. Sometimes pseudocyst are present in jaw bone. The absence of an epithelial lining precludes its classification as a true cyst [4]. Such pseudo cyst is rare in soft tissue of oral and maxillofacial region, we could not find any case so far. It is more common in pancreas. Pancreatic pseudocyst consists of localized fluid collection that is rich in pancreatic enzymes like amylase and is surrounded by a wall of fibrous tissue which is not lined by epithelial cells [5]. Alcohol-related pancreatitis is the major cause of pseudocyst of pancreas in countries where alcohol

consumption is high and it accounts for 59%–78% of all pseudocysts [6]. Non pancreatic pseudo cyst is rare occurrence and may be found in retroperitoneal region [7,8]. Treatment of the such lesion is enucleation. This rare case of Non pancreatic pseudo cyst is presented here according to SCARE 2020 guidelines [9]. This case was managed at Nepalese Army Institute of Health Sciences, college of Medicine, tertiary center, Kathmandu, Nepal.

## 2. Presentation of case

A 4-year-boy presented with one year history of a slowly growing mass in the right cheek. The patient had no history of pain, tenderness, dysphagia, dysphonia, dyspnea or any trauma on the face. There was no any relevant drug history, family history, psychosocial history or any relevant genetic information. Physical examination revealed a 3x2x1 cc mass on right cheek extending from the right commissure of mouth to the pterygomandibular raphe. The swelling was firm, non-tender, non-fluctuant, non-pulsatile, overlying mucosa was normal in colour, margin was distinct, freely mobile between the buccal mucosa and skin.

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Aspiration was negative. Submandibular or submental lymph nodes were not palpable. The mass was identified by ultrasonography. A computed tomography (CT) scan demonstrated a well-defined and hypodense soft tissue mass in the muscle layer of the right cheek (Fig. 1). The hemogram of the patient was within normal limits.

### 3. Surgical procedure

The surgery was performed by Dr. Manoj Adhikari, Oral and Maxillofacial Surgeon, under general anesthesia with the patient in supine position. Horizontal incision was made through the mucosa on the middle portion of the cyst. Careful blunt dissection was performed by scissors through submucosal tissue and buccinator muscle fibers. Then the cyst wall was visible and a hemostat was used to bluntly dissect the cyst from the surrounding subcutaneous tissue throughout the whole process to avoid damage to muscles, arteries, and nerves (Figs. 2 and 3). Simultaneously, the cyst was jacked with the other hand on the skin surface to provide better visibility, and was completely removed through the mucosal incision. The operated site was cleaned with povidone iodine – saline solution and was sutured with non-resorbable 3-0 silk sutures. All the post-surgical instructions were given to the patient and analgesics along with antibiotics were prescribed. The tissue specimen was dipped in 10% buffered formalin and sent to the histopathology lab for further examination. After one week, the sutures were removed and healing of the excised areas was found to be satisfactory and patient was recalled after three months, six months and one year for observation to prevent recurrence. No sign of recurrence was observed till one year follow up period.

#### 3.1. Intraoperative findings

Gross specimen of size 3x2x1 cc was excised. It was filled with clotted blood and was lined with fibrous tissue.

#### 3.2. Histopathological findings

Cyst shows no epithelial lining. A layer of hemorrhage and fibrous wall are seen. No atypia or evidence of malignancy is seen (Figs. 4 and 5).

### 4. Discussion

It is the first case of non-pancreatic pseudocyst found in soft tissue of oral and maxillofacial region. Non pancreatic pseudocyst is rare and may be present in retroperitoneal region [7,8]. The etiopathogenesis of these pseudocysts is not known yet. It may be considered as soft tissue

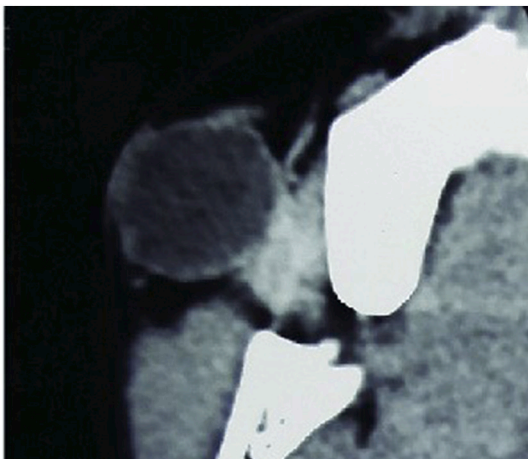


Fig. 1. CT scan of right cheek showing well defined hypodense lesion.

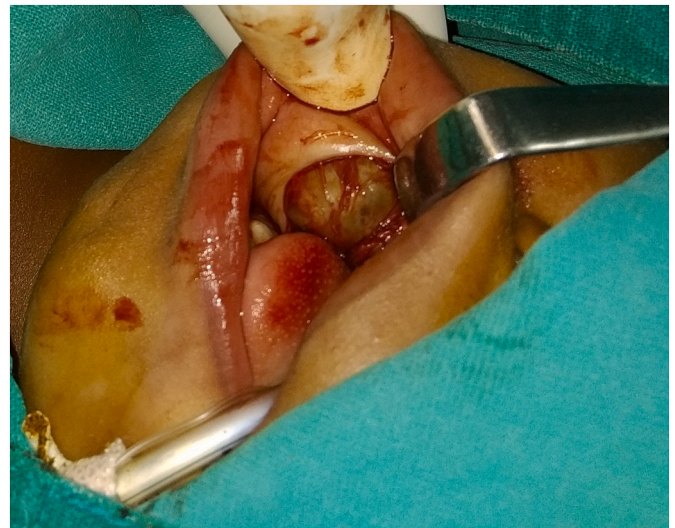


Fig. 2. Intact non-pancreatic pseudo cyst exposed on right cheek through buccal horizontal incision.

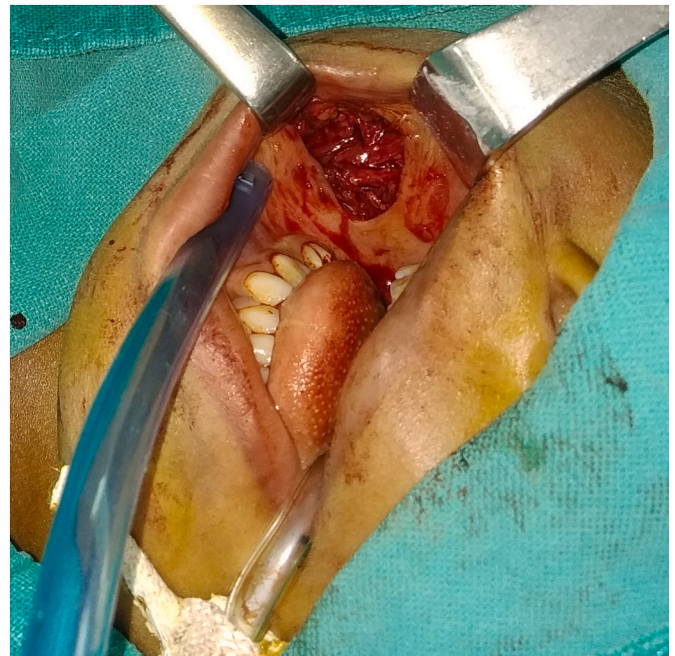
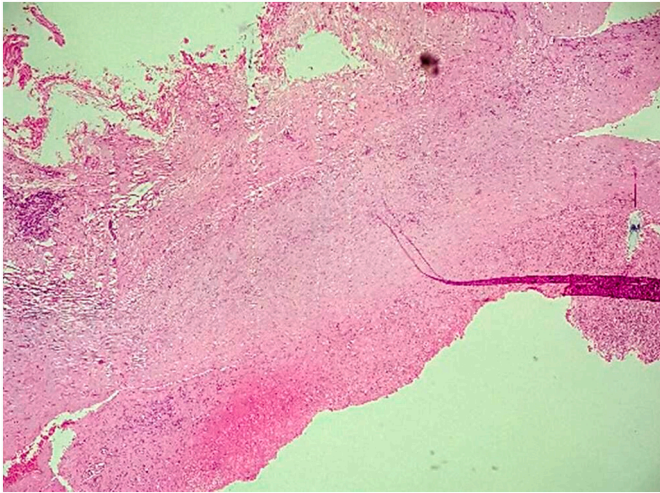


Fig. 3. Muscles bed seen after enucleation of non-pancreatic pseudo cyst from right cheek.

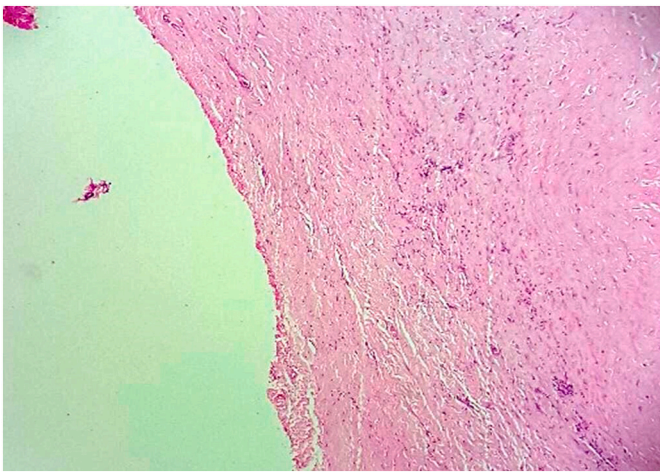
counterpart of Aneurysmal Bone Cyst (ABC). In case of ABC, Lichtenstein [10] proposed that alterations in local blood flow and increased venous pressure, and thus leading to engorgement of the vascular bed in the transformed bone, which leads to bone resorption, connective tissue replacement, and osteoid formation. It is also proposed that the lesion may represent an attempt to repair haematoma with the giant cell granuloma, but maintaining vascularity intact. Similar phenomenon may happen in formation of non-pancreatic pseudocyst in soft tissue of oral cavity.

In diagnosing such a rare lesion the clinicians may have broad variety of differential diagnosis. It can be classified as infectious, developmental and neoplastic processes.

The common infections in oral and maxillofacial region is odontogenic infections, like buccal space infection, masseteric space infection,



**Fig. 4.** 10x view, sections from a cyst shows a tissue with no definitive epithelial lining.



**Fig. 5.** 40x view, sections from a cyst shows a tissue with no definitive epithelial lining.

are unlikely in this case because the lesion has attained considerable size without any signs of infection/inflammation like malaise or fever.

Mucocele or ranula formation is another possibility. Ranula has characteristic bluish-gray hue which was not present in this lesion, the lesion had normal overlying mucosal color.

Malignancy is another possibility but owing to lack of nodal involvement, lesion's size, normal overlying mucosa, cystic homogeneity, this possibility was excluded.

Benign lesion in this region may include lipid, salivary, and vascular lesions. The appearance of lipoma is yellowish and nodular. A vascular lesion like hemangioma or lymphangioma have irregular mucosal surface and are lobulated. Hemangioma is reddish purple in color and lymphangioma is clear and translucent. This lesion did not have such features hence this diagnosis is also unlikely.

Most salivary gland tumors would cause an obstructive phenomenon when they reach this size. There were no any such features so this diagnosis was also excluded.

In case of cystic hygroma, it should be present at birth in 50% of cases or develop by two years of age in 90% of cases. It is commonly present in posterior triangle of the neck and are soft, fluctuant masses. In oral cavity it is present in anterior two third of tongue.

Another differential diagnosis may be oral lymphoepithelial cyst. It is

an uncommon within oral lymphoid tissue. It may be present anywhere within or adjacent to Waldeyer's ring. Its size is less than one cm, smooth, whitish-yellow, firm, painless mass. It usually contains cheesy keratinaceous material in the lumen. Due to the size, location, color, this diagnosis was also excluded.

## 5. Conclusion

Non pancreatic pseudo cysts may form in soft tissue of oral and maxillofacial region. They may be considered as soft tissue counterpart of Aneurysmal bone cyst of jaw bone.

The proposed treatment of pseudocysts in soft tissue of oral and maxillofacial region is surgical enucleation. The cyst removal procedure is simple and effective, and its success was confirmed by the lack of postsurgical alterations and no recurrence of the lesion.

## Patient perspective

The patient's parents were happy and grateful to the operating team.

## Declaration of competing interest

The authors declare that there is no conflict of interests regarding the publication of this paper.

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## Ethical approval

It is our routine standard surgical procedure so ethical clearance was not required.

## Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

## Author contribution

Manoj Adhikari: First Author and Corresponding Author.

Contributions: study concept or design, data collection, data analysis or interpretation, writing the paper etc.

Kanistika Jha: Co-Author.

Contributions: study concept or design, data collection, data analysis or interpretation, writing the paper etc.

## Research registration

This case report does not include any 'first in man' studies, so, registration was not required.

## Guarantor

Dr. Manoj Adhikari, Consultant Oral and Maxillofacial Surgeon, Nepalese Army Institute of Health Sciences, College of Medicine,

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*Provenance and peer review*

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