ORIGINAL RESEARCH PAPER
Volume - 12 | Issue - 02 | February - 2023 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

PHARYNGEAL ARCH CYST OF PAROTID- A CASE REPORT

Dentistry

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ABSTRACT

Branchial cleft cysts otherwise known as benign cervical lymphoepithelial cysts are the most common congenital neck masses, present in the lateral aspect of the neck. They originate from remnants of the branchial arches or branchial pouches. They most commonly occur in the angle of the mandible, in the submandibular area and parotid areas. In this article, we present a case of brachial cleft cyst of left parotid region in a 50year old female patient who reported with the complaint of slow growing painless swelling in the left pre auricular region and was diagnosed with brachial cleft cyst with further investigations and treated accordingly.

KEYWORDS

Brachial cleft cyst, brachial arches, congenital, asymptomatic.

INTRODUCTION

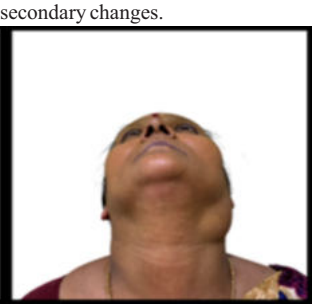
Branchial cleft cysts was first described by Hunczovsky in 1785. Branchial cleft anomalies arise when there is persistence of branchial cleft or pouch[1]. They are the most common congenital defects in head and neck region. 95% of these cysts arise from the second branchial arch and 5% originating from first, third and fourth branchial arches[2]. These are the most common congenital neck masses. It is also called as lateral lympho epithelial cyst, hygroma colli and Congenital hydrocoele of the neck. They usually present as fistulas, cysts and sinuses[3].

CASE REPORT

A 50 year old female patient reported with the chief complaint of swelling in the left lower third of the face below the left ear for past 1 year .Patient gave history of swelling, which was initially small, later increased in size gradually to attain the present size with no associated pain. Patient also gave history of dryness of the mouth. Her medical history was non-contributory. Her past dental history revealed uneventful extraction of upper right back tooth before one year. Her habit history revealed, she had the habit of chewing smokeless tobacco -2- 3 times/day for past 6 months and used to keep the quid in the left inner cheek region for 10 minutes and spits it off ,patient quit the habit before 1 month.

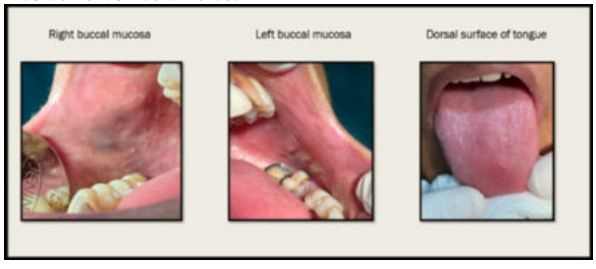
On extra oral examination (Figure-1,2), marked asymmetry of the right side of the face seen. Presence of single well defined oval shaped swelling in the left lower third of the face and in the left neck region measuring approximately 5*4cm in its greatest dimension extending superiorly below the left lobule of the ear to 2 cm below lower border of the mandible at the level of first neck crease line, anteriorly about 3cm from lip commissure to posteriorly 1cm from ramus of the mandible, just behind the left ear lobule. The skin over the swelling appears normal in accordance with adjacent skin and no secondary changes seen. On palpation, the swelling is firm in consistency, no warmth present, non-tender, non-compressible, non-reducible, freely mobile with no pulsations and no secondary changes.

Lateral View Left Side (Figure -1)



Worm's Eye View (Figure-2)

On intra oral examination (Figure-3), reduced mouth opening noted of about 26 mm. Reduced salivary flow on milking right and left stenson's duct, submandibular and sublingual gland salivary flow was normal. Presence of diffuse blanching of right and left buccal mucosa, floor of the mouth. No sinus opening or pus discharge (No secondary changes present). On palpation, vertical fibrous bands palpable on right and left buccal mucosa. Circum oral bands palpable. Tongue movements such as protrusion, lateral movements are normal. The dorsal surface of the tongue is smooth with areas of de-papillation. Uvula and palate normal. On hard tissue examination, root stumps in relation to 17,26,27, 36,38; dental caries in relation to 46,31,41; fractured 37; cervical abrasion in 44. On soft tissue examination, grade IV oral submucous fibrosis, generalized chronic gingivitis with localized periodontitis in relation to 31,32,33,41,42 was noted. With the patient's history and clinical findings, a provisional diagnosis of benign salivary gland neoplasm of left parotid was given. The most relevant differential diagnosis includes pleomorphic adenoma, Warthin's tumour, nodal metastatic tumour was given. Further patient was subjected to chair side investigation of facial nerve evaluation, which revealed wrinkling of forehead, eyes opening, closing, nasolabial fold are normal. Patient is able to smile and puff her cheek completely, normal muscle activity in relation to orbicularis oris, orbicularis oculi, buccinator and platysma muscles, which finally indicated no involvement of facial nerve.



INTRA ORAL (FIGURE-3)

INVESTIGATIONS DONE

Patient was further subjected to radiographic investigations,

1) **OPG revealed (Figure-4)**

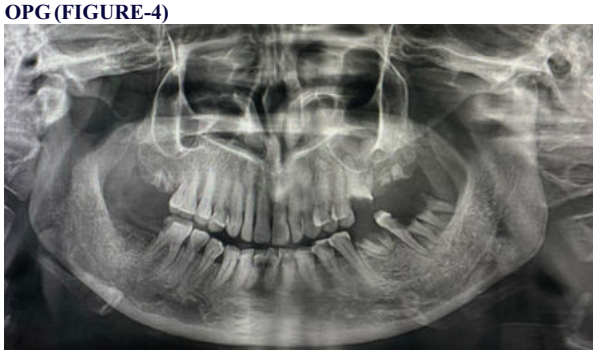
Zone -1

No of teeth -28
Missing- 18,28,47,48
Root stumps- 17,26,27,36,38
Coronal radiolucency involving enamel, dentin, pulp extending till furcation region with loss of distal portion of crown structure – Fractured and Dental caries 37
Mesio-proximal radiolucency involving enamel, dentin, pulp with

widening of PDL space in relation to 31,41- DC with apical periodontitis 31,41
Disto-proximal radiolucency involving enamel, dentin suggestive of Dental caries -46

Zone -2,3,4,5,6
Nose, Maxillary sinus, angle, body and ramus of mandible, inferior border of mandible, TMJ, hyoid bone normal.

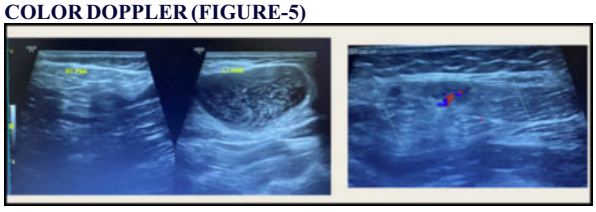
OPG (FIGURE-4)



2) **Ultrasound Of Right And Left Parotid (Figure-5)**
In left parotid -A well-defined thin-walled cyst of size 4.8*3.4*2.4cm with hyperechoic contents showing brownian motion within, is seen in the inferior half of the superficial lobe of left parotid gland. Few sub centimetric lymph nodes seen adjacent to left submandibular gland (Level Ib). In right parotid -It appears normal in size, shows homogeneous echoes. No focal lesion seen. No ductal dilatation seen. No calculus seen.

Impression- Left parotid abscess

ULTRASOUND OF RIGHT AND LEFT PAROTID WITH COLOR DOPPLER (FIGURE-5)



3) **ULTRASOUND GUIDED FNAC revealed yellowish viscous fluid (Figure-6)**
Microscopic Features
Negative for malignant cells. Smear show acellular proteinaceous material admixed with scattered cyst macrophages and few inflammatory cells.

Impression: Features are that of a benign cystic lesion – MILAN category -II

Differentials include-
1) Retention cyst
2) Lymphoepithelial cyst

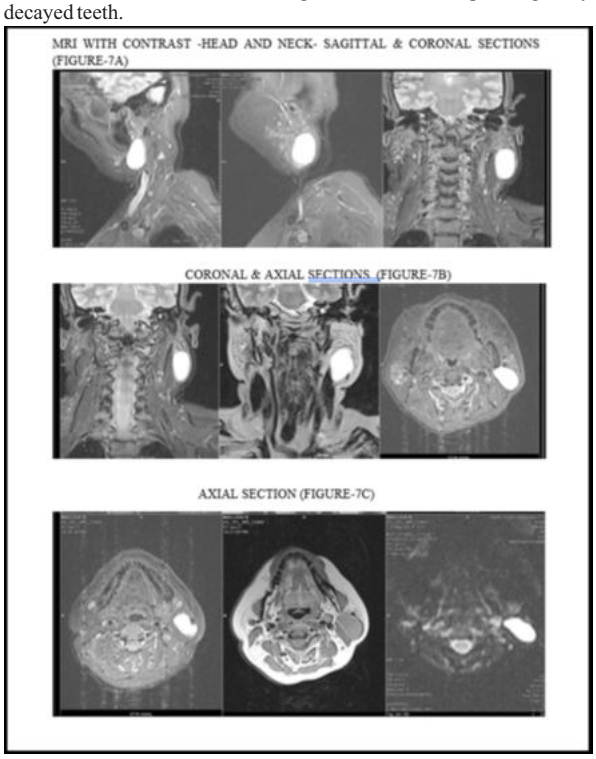
ULTRASOUND GUIDED -FINAL NEEDLE ASPIRATION CYTOLOGY- LEFT PAROTID (FIGURE-6)

4) **MRI PAROTID GLANDS:** T2 FS Axial, Coronal and Sagittal (Figure -7A,7B,7C)

T2W Coronal, T1W Axial, CISS Sequence revealed; Well-defined T2W hyperintense and T1W hypointense cystic lesion with facilitated diffusion seen in the inferior aspect of left parotid gland at the level angle of mandible and displacing the external jugular vein laterally-suggestive of Benign intra parotid cystic lesion (Possibilities-Branchial cleft cyst, Benign lymphoepithelial cyst). Few cervical lymph nodes are seen in the bilateral level I and II (Reactive).

Patient was further subjected to blood investigations and serological workup, which revealed, she was found negative for HIV-I, II; HBsAg; HCV and her complete blood count, blood glucose level and urine culture was found to be normal. A final diagnosis of branchial cleft cyst of left parotid was given based on the patient's history, clinical and radiographical investigations. Patient was further subjected to surgery under GA for removal of the cyst.

Patient was prescribed with capsule SM FIBRO (1-0-0) -30 days for management of oral submucous fibrosis and asked to report after a month for review and for the management of root stumps and grossly decayed teeth.



DISCUSSION
Patient presenting with neck lump can lead to lot of differential diagnosis. When the lump is superficial to underlying muscle and fascia, sebaceous cyst or lipoma can be considered[4]. But if it is present deep, diagnosis can be made based on clinical history and physical examination. They can be either inflammatory, congenital, neoplastic and others (sialolithiasis). In inflammatory, they can be either due to bacterial (streptococcal, staphylococcal), viral (Epstein Barr, HIV, Cytomegalovirus) or due to protozoal (Toxoplasmosis, Leishmaniasis). Autoimmune diseases include sarcoidosis, lupus and Sjogren's syndrome[5]. In congenital diseases, branchial cleft cyst, hemangioma or lymphangioma can be considered if it is asymptomatic and present for months or years. In neoplastic category, we have benign tumors such as pleomorphic adenoma, schwannoma, paraganglioma, neurofibroma or thyroid tumors. Malignant tumors such as metastatic Head and Neck Squamous cell carcinoma (considering HPV etiology, young male patient), other metastatic tumors (considering lung, breast, upper GI primary)[6].

Considering the salivary glands (ie., parotid gland), the differential for cystic parotid lesions includes:
Bilateral cystic parotid lesions such as warthin's tumor, benign lymphoepithelial lesions of HIV, Sjogrens's syndrome and sialoceles. Unilateral cystic parotid lesions includes warthin's tumor, sialocele, first branchial cleft cyst, parotid lymphangioma, benign lymphoepithelial cyst, necrotic lymph nodes.

Branchial cleft anomalies are congenital defects that occur in the head and neck. The anomalies result from branchial apparatus (six arches; five clefts), which are the embryologic precursors of the ear and the muscles, blood vessels, bones, cartilage, and mucosallining of the face, neck, and pharynx. During the 3rd to 5th week of embryonic

development, the second arch grows caudally and covers the third, fourth and sixth arches. When it fuses to the skin caudal to these arches, the cervical sinus is formed. Eventually, the edges of cervical sinus fuse and the ectoderm within the tube disappears. Persistence of branchial cleft or pouch results in a cervical anomaly located along the anterior border of the sternocleidomastoid muscle from the tragus of the ear to the clavicle[8]. First branchial cleft anomalies seen above the level of the mandible near the external auditory canal within or close to the parotid gland. Second branchial cleft anomalies seen between the level of the mandible angle and the carotid bifurcation, deeper than the platysma and superficial layer of deep cervical fascia. Third branchial cleft anomalies are rare in infrahyoid neck. Fourth branchial cleft anomalies are rare and occurs adjacent to the thyroid gland. The fifth cleft does not give rise to the cervical sinus of HIS which is part of the reason that there are no fifth branchial cleft anomalies. Many theories have been proposed for origin and development of branchial cysts, which are Branchial apparatus theory, cervical sinus theory, thymus-pharyngeal duct theory and inclusion theory[9].

First branchial cleft cysts are uncommon and represent only ~7% of all branchial cleft cysts, occur in middle-aged women. Its clinical presentation includes asymptomatic, palpable lump or inflammatory mass in the parotid region, spontaneous fluid draining from a pit-like depression on the skin, which may be mucus or pus depending on the presence of associated infection. Facial nerve palsy can occur. First branchial cleft cysts develop as a result of the incomplete fusion of the cleft between the first and second branchial arches. They typically occur within or close to the parotid gland or external auditory canal. Branchio-Oto renal and Branchio-Oculo facial syndrome are syndromes associated with it. Its subtypes include; Type I: Infero-posteromedial to the pinna; Type II: Between the angle of the mandible and the external auditory canal; Type IV: Peri-parotid. Histologically it can be classified into two types[10].

Second branchial anomalies are the most common and represent more than 90% of all branchial cleft cysts. They communicate with oropharynx via supra-tonsillar fossa. Cysts are more common than sinus and fistula are the least common[11]. Bailey classified second branchial cysts into four types;
Type-I : Superficial cysts lying anterior and adjacent to sternocleidomastoid muscle
Type-II : Cysts lying on greater vessels, may adhere to internal jugular vein
Type-III : Lesions extending between internal and external carotid arteries
Type-IV : Lesions lying in the parapharyngeal space next to pharyngeal wall.

Investigations are mandatory for diagnosis of these lesions. Fine needle aspiration biopsy, Ultrasound, CT and MRI provide clear image of the lesion and reveal intra-parotid cystic masses. MRI is the imaging modality of choice and helps in characterizing the lesion. The definitive diagnosis of the cystic lesion of the parotid gland depends solely on Histopathological examination (HPE) as also in our case.

Diagnostic categories and risk of malignancy in the Milan system of reporting salivary gland cytopathology includes six categories and their risk percentage of turning into malignancy (Table-1). Serological testing for HIV-I, II, Hepatitis is very important to rule the viral etiology as these cysts are more common among HIV patients. The differential diagnosis of this cyst includes Warthin's tumor, intramuscular benign hemangioma (IMH), branchial cleft cyst and lymphoma. Treatment mostly involves complete excision and relapse rate is less in branchial cleft cyst cases. Branchial cleft cysts have neither been known to recur nor metastasize. For this reason, it is important that they should be differentiated from aggressive lesions, especially cystic low-grade mucoepidermoid carcinoma.

MILAN SYSTEM OF REPORTING SALIVARY GLAND CYTOPATHOLOGY (TABLE-1)

DIAGNOSTIC CATEGORY	RISK OF MALIGNANCY (IN %)
1) Non -diagnostic	25%
2) Non-neoplastic	10%
3) Atypia of undetermined significance (AUS)	20%
4) Neoplasm 4a) Neoplasm : Benign 4b) Neoplasm : Salivary gland neoplasm of uncertain malignant potential (SUMP)	<5% 35%
5) Suspicious for malignancy (SM)	60%
6) Malignant	90%

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