**UNCOMMON ORIGIN OF PAROTID SWELLING**

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**Abstract**

First branchial cleft cyst is a rare disease of the head and neck. A branchial cleft cyst commonly presents as a unilateral solitary, painless soft tissue swelling in the upper region in the neck. It is clinically apparent in late childhood or early adulthood. First branchial cleft cyst accounts for around 8% for all branchial abnormalities. It is possible for first branchial cleft cyst to be easily misdiagnosed as other swellings of the head and neck region. It is imperative that clinicians to make an accurate diagnosis so that appropriate treatment can be offered. A case report of type I first branchial cleft cyst is presented here.

**Keywords:** First branchial cleft cyst, Cervical lymphoepithelial cyst, Parotid gland

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**CASE REPORT**

**Introduction**

First branchial cleft anomalies (FBCA) is a developmental anomaly thought to originate from the branchial apparatus that did not completely obliterated during head and neck embryogenesis [1]. FBCA although relatively rare, contribute around 8% for all branchial anomalies. The incidence is estimated to be about one per million population per year [2]. Female are twice as frequently affected than male with a tendency to occur on the left side. The spectrum of developmental abnormalities includes cysts, sinuses fistulas and various combinations of these entities.

FBCA are due to incomplete fusion of ventral portion of the first and second arches. During development, the closure time of the cleft is earlier in comparison with facial nerve and parotid gland, thus FBCA have a close relationship between these structures. As obliteration of the cleft proceeds from ventral to dorsal, the lesion often occurs near the ear and parotid gland. FBCA can also occur in any part of the external auditory canal. Many FBCA are asymptomatic however they may become enlarged or inflamed especially during periods of upper respiratory tract infection owing to the lymphoid tissue located beneath the epithelium. This is usually the first instance that the swelling is realised. The principle of management includes early and accurate diagnosis, controlling infection status and complete excision without facial nerve injury. Prognosis are generally good for patients.

**Case Report**

A 11-year-old female presented with a chief complaint of sudden increasing swelling over the left parotid region for three months. The swelling increased in size to 6 x 5 cm from a small 2 x 2 cm with difficulty in mastication. In spite of the enormous size, there were no associated complaints such as change in voice, breathing difficulty, facial asymmetry, ear discharge or episodes of infection.

On examination the patient was healthy, afebrile and well nourished. An ovoid solitary firm swelling was observed over the left parotid region without overlying skin changes. On palpation it was non tender and non-pulsatile. The swelling was also attached to the underlying structures. Examination of the nose, ear and oral cavity demonstrated no abnormality. A flexible nasopharyngoscopy was performed showing normal findings.

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A fine needle aspiration cytology had aspirated three millilitres of cloudy fluid. Subsequently, she underwent a contrast enhanced computed tomography (CECT) examination of the neck which showed presence of a well-defined cystic mass with mildly thickened enhancing wall located in the superficial lobe of the left parotid gland. The cystic mass measured 2.8 x 3.3 x 4.7 cm. Thin enhancing septae were also seen within the mass. There were no enhancing nodules, layering of densities or calcifications seen in the mass. The mass was confined within the parotid gland. It appeared to be compressing the left masseter muscle. Thyroid gland, pharynx and laryngeal structures were normal. There was no evidence of lymphadenopathy. Vascular structures in the neck were normal in course, calibre and enhancement pattern. The facial nerve was not able to be seen in the study. No other abnormalities were detected (Figure 1).

![Contrasted CT scan of the neck in axial (a), coronal (b) and sagittal (c) planes showing presence of a well-defined, cystic mass (CM) with mildly thickened enhancing wall in the left parotid gland (P). It causes mass effect onto the masseter muscle (M) anteriorly. SG (submandibular gland), SCM (sternocleidomastoid muscle), EJV (external jugular vein).](image1)

**Figure 1.** Contrasted CT scan of the neck in axial (a), coronal (b) and sagittal (c) planes showing presence of a well-defined, cystic mass (CM) with mildly thickened enhancing wall in the left parotid gland (P). It causes mass effect onto the masseter muscle (M) anteriorly. SG (submandibular gland), SCM (sternocleidomastoid muscle), EJV (external jugular vein).

![Photomicrograph showing fibro-collagenous cyst wall lined by flattened stratified squamous epithelium to ciliated pseudostratified columnar epithelium associated with lymphoid component within the cyst wall. Yellow arrow showing normal parotid gland.](image2)

**Figure 2.** Photomicrograph showing fibro-collagenous cyst wall lined by flattened stratified squamous epithelium to ciliated pseudostratified columnar epithelium associated with lymphoid component within the cyst wall. Yellow arrow showing normal parotid gland. (H&E stain, X 40).
Left parotidectomy was done and intraoperatively the lesion was seen to be arising from the deep lobe of parotid. Thus, both lobes of parotid had been removed with preservation of the facial nerve and its branches. The patient developed a left facial asymmetry (House Brackmann grade IV) post operatively. Artificial eye drops and patch with regular facial exercise had been initiated early during the stay. She was subsequently discharged three days after the surgery. Histopathological examination revealed uniloculated cyst with fibrous cyst wall mainly lined by respiratory-type epithelium and focally lined by non-keratinized stratified squamous epithelium and it’s consistent with benign lymphoepithelial cyst (Figure 2 and 3). She was seen back in the clinic two weeks after with good improvement of her facial weakness and good wound healing. The patient was followed up for a year with residual facial asymmetry (House Brackmann grade II) and no evidence of recurrence was observed.

Discussion
Branchial cleft cyst or known as cervical lymphoepithelial cyst is a common cause of soft tissue swelling in the neck region of young adults. They generally occur unilaterally and are typically seen in the lateral aspect of the neck [1]. Differential diagnosis such as congenital hydrocele of neck, hygroma colli and lymphangioma should be considered in younger children whereas in older adults it is important to exclude metastatic lymphadenopathy, lymphoma and tuberculosis.

First branchial cleft anomalies (FBCA) are uncommon that arise from incomplete closure of ventral portion of the first branchial cleft [2]. Initially completes its development into external ear canal, muscles of mastication, maxilla, mandible and portions of middle ear structures (malleus and incus) by the sixth and seventh weeks. As the parotid gland and facial nerve develop later, a FBCA is located around them. Thus, the first branchial cyst can originate anywhere along the nasopharynx, middle ear cavity or external auditory canal. They may also involve the parotid gland or lie medially or superficially to it and also true for facial nerve. This type of abnormality comprises around 8% of all incidence of branchial cleft anomalies. There had been various attempts to classify FBCA. The most widely accepted theory was proposed by Arnot in 1971 [3]. A type I lesion was defined as any cyst or sinus in the parotid gland lined by squamous epithelium whereas type II lesion was defined as cyst of sinus that communicate with the external auditory canal. These abnormalities according to Arnot resulted from incomplete obliteration of the cleft. In our case, the swelling is arising from the parotid region which is uncommon for branchial cleft cyst. No history of persistent ear discharge as it might show a possibility of communication with the external ear canal. Considering the embryogenic origin, the possibility of the cyst in our case might be originating from the first branchial cleft.

The diagnosis of FBCA is made primarily using medical history, clinical manifestation and exclusion as for other conditions. Pre-operative diagnostic procedures include computed
tomography (CT) or magnetic resonance imaging (MRI) scan and fine needle aspiration. With imaging it does not only confirms the cystic nature of branchial anomaly, but also determines the extend and anatomical relationship with adjacent structures such as external carotid artery, retromandibular vein and facial nerve. These information are vital for surgical planning and preservation. MRI is more superior to locate and identify soft tissues such as nerve. Reports in the literature suggest that FBCA is superficial in relation with the facial nerve in majority of cases. According to Arturo et al case series, first branchial anomaly is spread evenly in comparison with the facial nerve [4]. The index patient underwent a standard parotidectomy using a facial nerve monitor. All main branches of facial nerve tributaries were anatomically and physiologically identified and preserved. She developed temporary facial asymmetry post operatively. There is 21% incidence of temporary facial nerve palsy in patients in whom the nerve was identified and 29% in patients in whom the facial nerve was not identified [5].

Surgical excision is still the definitive treatment for FBCA. Understanding of various specific types of lesions and their relationship with the surrounding structures example facial nerve is paramount. Successful surgery mandates complete resection and facial nerve preservation. Other complications of surgery include formation of persistent fistula and recurrence are very rare.

**Conclusion**
The uncommon first branchial cleft cyst should always be one of the differentials for neck swelling including the parotid region. Though it can present in various ways a good history taking and examination should be done. It shares a clinical presentation with other pathological entities of the neck such as infection such as mumps, reactive lymphadenopathy and even malignancy; thus detailed history taking and examination had to be done for every patient that presenting with head and neck swelling. Surgery is still the main stay of treatment for FBCA with facial nerve preservation. There will be multiple and various presentations and relationship with facial nerve. Thus, imaging will be just a guide for the operative surgeon. Good understanding of cyst with its surrounding structures are the key of successful surgery. Successful surgery mandates complete resection and facial nerve preservation.

**References**


