CASE REPORT

Parotid Lymphoepithelial Cyst: A Case Report
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ABSTRACT
A 48 years old Malay lady with a case of painless soft fluctuant swelling of left parotid gland is reported. The lesion was found to be a cystic lesion through the pre operative examinations and investigations. The cyst was completely excised, taking care not to injure the lower division of the facial nerve. Post recovery was uneventful with no defect of the facial nerve functions. The histologic picture confirmed that the cyst was lymphoepithelial cyst which is so called “branchial cyst”. Through the literature reviews of parotid lymphoepitelial cyst the discussions on prevalence, origin, diagnosis, histological finding, investigation and the modes of treatment are made. The ultra sound was found to be valuable in the pre operative evaluation of the parotid swelling furthermore it is non-invasive, harmless, painless and relatively quick.

Keywords: branchial cyst, lymphoepithelial cyst

INTRODUCTION
Cystic lesions within the parotid gland are uncommon, comprising approximately 5% of all salivary gland tumours, many of them represent cystic components of neoplasms (Altman and Bailey, 1994). Lymphoepithelial (so-called branchial) cysts are most often found in the lateral cervical area just below the angle of the mandible, anterior to the sternocleidomastoid muscle. However, sometimes they may occur in the parotid gland (Fujibayashi and Itoh, 1981). The cysts have equal distributions among males and females, and usually present as a painless swelling in the parotid area without attachment to the facial nerve (Antoniadis et al., 1990). The true origin of this cyst which thought to be congenital remained unclear, however several theories/hypothesis have been proposed such as classic branchial cleft theory (Rickles and Little, 1967), parotid gland inclusion theory (Bashkar and Bernier, 1959) and thymopharyngeal duct theory which was proposed by Wenglowski (1913) and later supported by Meyer and McNealy (Camilleri and Lloyd, 1990). However, in the last few years, such cysts have been found in increasing numbers in AIDS patients as well as in the patients belonging to the AIDS-risk groups (Marmary, 1990). While the etiology of these cysts is controversial the treatment is not. Many authors suggested for complete excision with sufficient margin of normal tissue surrounding. However the diagnosis is seldom made pre-operatively therefore superficial parotidectomy is another form of treatment (Maynard, 1988; Wayman et al., 1988).

REPORT OF CASE
A 48 years old Malay lady with the big swelling in the left parotid region was referred to Oral & Maxillofacial Department of Kota Bharu Hospital from Kuala Balah Rural Health Center on Mac, 2004. She started to notice this swelling somewhere in Dec, 2003. Since then it gradually increase in size, however it was entirely asymptomatic. She denied any history of upper respiratory tract infection or trauma to the parotid area. Generally, she was stable and afebrile. Her general medical history was hypertension which is controlled with metoprolol, 100 mg and nifidipine, 10 mg. From the family history she was a rubber tapper and a part time religious teacher with five children and a grandchild.

On extra oral examination revealed that 6 cm x 6 cm diffused soft fluctuant swelling in the left pre and inferior auricular area with elevation of lower ear lobe. The overlying skin noted to be normal in colour and freely moveable over the mass. The function of facial nerve was normal. The swelling appeared to be fluid filled, with no audible bruit was detected. There was no associated regional lymphadenopathy or thyroid enlargement. Intra oral examination revealed a free flow of saliva from the left parotid (Stensen’s duct) opening.
Further investigations on her were done including plain radiograph of orthopantomographic radiograph (OPG) and lateral skull, however both revealed normal conditions. Ultra sound was then performed at Radiology Department and the report was a hypoechoic with posterior enhancement which suggestive for cystic swelling. The content of the cyst aspirated to look for the nature of the fluid, in which was found to be a straw colour containing whitish cheesy materials. The aspirate was then sent to the Pathology Department for cytology examination. The result was favour with no malignancy cells and again suggestion of parotid cyst was made. Further investigation with MRI to locate the extent and relation of lesion was not done as the patient refused to go to the other hospital as it was not available in our hospital. Based on clinical history and presentation, aspiration and ultra sound, the pre operative diagnosis of parotid cyst was made.

The finding was explained and patient agreed for the surgical excision under general anaesthesia. On the routine pre operative assessment she was found to have high random blood sugar level > 18.3 mmol/dl and urine analysis confirmed the presence of glucose in the urine. Incidentally she was diagnosed to have diabetes mellitus. She was then started on DK regime for the sugar correction. Her blood pressure

Figure 1. Front view of the parotid cyst

Figure 2. Lateral view of the parotid cyst

Figure 3. Orthopantomogram findings
was controlled with regular medications. At induction, the prophylaxis IV Cefuroxime 1.5 gm was given together with IV Dexamethasone 8 mg. After skin preparation with povidone solution, assess to the cyst was made through the lower neck incision at the deepest neck crest which is found to be posteroinferior to the cyst.

This will help to minimise the post operation scarring and avoiding from puncturing the cyst. The incision was then extended to the mastoid, posterior and inferior auricle. The incision was then performed layer by layer until it reached the platysma muscle. This muscle was dissected to exposed the underneath cyst capsule. Once the plane between cyst capsule and platysma muscle was identified the blunt dissection was carried out carefully separating the cyst from the sternocleidomastoid muscle posteriorly and the posterior belly of digastric muscle underneath with the preservation of facial nerve. However the upper part of the cyst was found embedded in the parotid tissue, so part of the parotid tail was excised together with the cyst. The specimen was then sent for histopathology examination. After the haemostasis was achieved, the surgical area was placed with Radivac drain and then closure was done in layers with 4O vicryl and 5O defilon. The patient recovered from surgery uneventful. The subcutaneous insulin was then changed to metformine 500 mg TDS and was discharged home next day. She was seen at Dental Clinic a week post surgery with no evidence of facial nerve palsy and wound infection.

The histopathology examination showed a collapsed cyst wall with adjacent salivary gland and small reactive lymphoid tissue. The cyst was lined by columnar epithelium with abundant, granular cytoplasm and densely eosinophilic (oncocytic/ oxophilic change). However no nuclear atypia was seen. The appearances were those of a lymphoepithelial cyst.

**DISCUSSION**

The definitive diagnosis of the cystic lesion of the parotid gland depends solely on histological examination (HPE). In this present case the diagnosis of parotid lymphoepitelial cyst was made after the HPE. Lymphoepitelial (so-called branchial) cysts within the parotid gland are rare. The first reported case of branchial cyst in the parotid gland was in 1895 by Hildebrant. Since then about 70 cases of this type of cysts have been reported (Camilleri and Lloyd, 1990). Another 33 cases was found by Fujibayashi through the reviewed of 5 publications which concerning of either branchial cyst or parotid diseases. The ages of patients ranged from 16 to 69 with the mean age of 44 years old. The distribution was found to be three times more frequent in males than in females. The majority of cases were unilateral with a greater number arising in the right parotid gland rather than the left gland (Fujibayashi and Itoh, 1981). The location of cyst in the present case was uncommon.

**Figure 4.** Operative view of the cyst
The most common sites for lymphoepitelial cysts were in the lateral cervical area. In the review of 468 cases of branchial cyst from the file of the Armed Forces Institute of Pathology only 5 cases were located in the parotid area (Bashkar and Bernier, 1959). On the other hand, a review of 149 cases of branchial cysts, reported that only 14 cases were found at upper part of the neck above the angle of the mandible (Rickles and Little, 1967). Many authors presented the similar clinical presentation of these cysts which appears to be slow growing painless swelling with normal movable overlying skin (Fujibayashi and Itoh, 1981; Skouteris et al., 1989; Altman and Bailey, 1994). The age and clinical symptoms of present case were found to be consistent with the literature reviews. The origin and development of branchial cysts is a controversial subject, and many theories have been suggested. The theories simply attempt to collate known embryological fact with histological and clinical findings. However to this day the controversy still exists and at least four theories have been put forward to explain the origins of branchial cysts, which are (Camilleri and Lloyd, 1990)

1) Branchial apparatus theory
2) Cervical sinus theory
3) Thymus-pharyngeal duct theory
4) Inclusion theory

The first two theories are also known as classic theory holds that the cysts develop from the remnants of the branchial cleft because it occurs in the area of the embryonic gill apparatus. However for this present case, the inclusion theory or so-called recent theory would seem the most feasible explanation for the lymphoepitelial cyst which was found in the parotid gland. Where this theory considers that the cysts arise from cystic changes in parotid gland epithelium that become entrapped in the upper cervical lymph nodes during embryonic life (Bashkar and Bernier, 1959). However, in the recent years the parotid lymphoepitelial cysts have been reported increasing in numbers and in close association with human immunodeficiency virus (HIV) infected as well as HIV-high risk patients. These are probably related to intraparotid lymphadenopathy associated with HIV infection (Mandel and Reich, 1992).

Presently, we have not conducted the HIV test on the patient as she is not belongs to the HIV-high risk group based on social and family history. The pre operative diagnosis of this case remains uncertain as the nature and clinical symptoms resemble the other cystic lesions of the parotid such as retention cysts, extravasation cysts or cystic degenerative salivary gland tumour. In which the latter will affect the choice of the treatment. The investigations are important in diagnosis and treatment planning of these lesions as the clinical examination may not always confirm the cystic nature of the lesions. In this present case, ultra sound scanning was found to be helpful in showing the cystic nature of lesions. Wyman et al. (1988) concluded that ultra sound scanning was a simple investigation that was recommended for pre operative diagnosis of cervical swelling. In which
the nature of echoes could be used to distinguish between cystic and solid tissue. Beside that, aspiration was particularly useful in identification of the nature of the cystic content. The aspirate was usually clear watery or straw colour fluid. Although it was not a sensitive investigation technique however further evaluation of the aspirate can provide additional information about the presence of cells, their type and morphology (Skouteris et al., 1989). In this case, magnetic resonance imaging (MRI) was not done as it was not available in our hospital. MRI could give clear images of the lesion and its anatomical relations, and it was generally more sensitive than CT scanning in detecting small intra-parotid masses (Altman and Bailey, 1994).

The histologic picture in this present case was in accordance with that of a lymphoepithelial cyst, where the epithelial-lined cysts were observed in a lymph node, adjacent to or embedded in a major salivary gland (Elliott and Oertel, 1990). In the review of literatures (Skouteris et al., 1989) reported that more than 90 percent of these cysts were lined by stratified squamous epithelium that might or might not be keratinized. Some cysts demonstrated respiratory epithelium. However there were reported cases where the cysts were lined by either columnar or cuboidal epithelium which was consistent with our present case.

Regardless of possible theories of origin, anatomy, histological and clinical behaviour which was discussed, clearly, many authors suggested total excision of these cysts were necessary to prevent infection and sinus formation (Maynard, 1988). However, the diagnosis was seldom made pre operatively therefore superficial parotidectomy was suggested in the case which the nature is doubtful during operation (Wyman et al., 1988). In present case the lesion was found well capsulated however the upper part of the cyst was stuck and embedded in the parotid tail tissues. The attempt to excise the part of parotid gland together with the cyst was made as suggested by some authors. The recurrence following complete surgical excision had not been reported. Rare examples of malignant transformation in these cysts had been reported. Although such an occurrence is theoretically possible, most of these cases represent cystic metastases from previously undetected carcinomas of the head and neck region, especially the nasopharynx.

REFERENCES


