

A Type II First Branchial Cleft Cyst Masquerading as An Infected Parotid Warthin's Tumor

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The diagnosis of a parotid mass usually depends on thorough history taking and physical examination. Diagnostic modalities, including ultrasonographic examinations, computed tomography and magnetic resonance images, may also provide substantial information but their accuracy for diagnosis is sometimes questionable, especially in differentiating some rare neoplasms. First branchial cleft cysts (FBCCs) are rare causes of parotid swelling and comprise less than 1% of all branchial anomalies. They are frequently misdiagnosed due to their rarity and unfamiliar clinical signs and symptoms. We present a case of type II FBCC masquerading as an infected parotid Warthin's tumor. We also review the clinical signs and symptoms of FBCCs in order to remind clinicians that this rare branchial anomaly can mimic an infected Warthin's tumor and may be seated in the deep lobe of the parotid gland. By making an accurate pre-operative diagnosis of type II FBCC, we can minimize surgical morbidity and avoid incomplete resection and possible recurrence. (*Chang Gung Med J* 2006;29:435-9)

Key words: first branchial cleft cyst, parotid mass, Warthin's tumor.

Sometimes the diagnosis and management of neck masses are a great challenge to the clinician even after thorough clinical evaluation. Warthin's tumor (papillary cystadenoma lymphomatosum) is the second most common benign neoplasm of the parotid gland, the first being pleomorphic adenoma.⁽¹⁾ The tumor predominately occurs in older men and usually presents as a slow growing mass in the parotid tail. However, branchial cleft cysts can also be congenital lesions manifesting in late childhood or young adulthood. They usually become apparent after upper respiratory tract infections.⁽²⁾ Infected cysts may develop into abscesses and further complicate into deep neck infection. First branchial cleft cysts (FBCCs) are rare and comprise less than 1% all branchial anomalies.⁽³⁾ Their embryological development remains unclear. Work classified FBCCs into

two types.⁽⁴⁾ Type I, which is ectodermal in origin, is considered to be a duplication of the membranous external auditory canal. It usually parallels the external ear canal and runs laterally to the facial nerve. Type II is of both ectodermal and mesodermal origin with cartilaginous components. It may pass medially or laterally to the facial nerve and rarely divides its main trunk. The diagnosis of FBCC, prior to surgery, is mostly based on clinical signs and symptoms. Although these, along with radiological images, are diagnostic, they can be easily misdiagnosed. We describe a case of type II FBCC in a 71-year-old man with presenting signs and symptoms of an infected Warthin's tumor. We aim to remind clinicians that this rare branchial anomaly could mimic an infected Warthin's tumor and be seated in the deep lobe of the parotid gland. Accurate pre-operative diagnosis of a

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type II FBCC is the cornerstone to avoiding unexpected morbidity and incomplete resection with resultant recurrence.

CASE REPORT

A 71-year-old man visited the otolaryngology clinic with a left neck swelling that had been present for one month and had rapidly increased in size over a 3-day period. On examination, a tender mass with unclear border was palpated in the infra-auricular area of the left neck. The lesion was about 7 × 8-cm in diameter, elastic, movable, painful, febrile and fluctuated. No other specific findings in the head and neck region were noted. Under suspicion of left acute suppurative parotiditis, the patient was hospitalized for antibiotic treatment. After 3-day intravenous antibiotics therapy, the mass decreased in size and febrile episodes subsided. Sonographic examination showed a cystic mass 2.7 × 1.6 × 1.5-cm in size in the left parotid area. Using echo-guided fine needle aspiration, 7-ml of yellowish fluid was drawn off and sent for cytological examination. The cytological examination showed polymorphonuclear cells, necrotic debris and squamous metaplasia with focal oncocytic cells, which suggested an inflamed Warthin's tumor. Contrast enhanced computed tomography (CT) scan demonstrated a 2 × 2-cm well-circumscribed neoplasm within the left parotid gland, which suggested an infected cystic mass (Fig. 1).

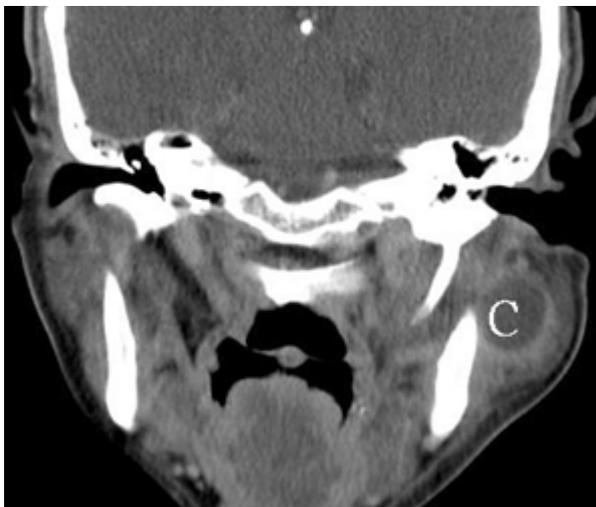


Fig. 1 Coronal view of computed tomography scan showing a cystic mass 2 × 2-cm in size in the parotid gland. C: cyst.

Under suspicion of Warthin's tumor, our initial surgical plan was a superficial parotidectomy with facial nerve dissection, which usually suffices for Warthin's tumor. However, during the operation, we found that the cystic mass was located medially to the facial nerve and extended toward the external auditory canal but did not communicate with it. After superficial parotidectomy (Fig. 2A), we gently retracted the buccal and marginal mandibular branch of the facial nerve and completely excised the 1.5 × 1.5-cm cystic neoplasm (Fig. 2B). The patient's post-operative course was uneventful; no facial nerve paralysis occurred. Pathological examination of the surgical specimen confirmed the diagnosis of FBCC (Figs. 3A, 3B). No sign of recurrence was noted after a follow-up period of 15 months.

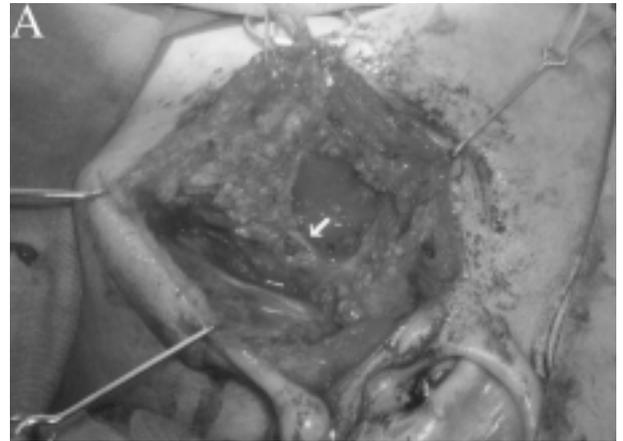


Fig. 2A Branches of the facial nerve exposed after performing a superficial parotidectomy. (Arrow: facial nerve)

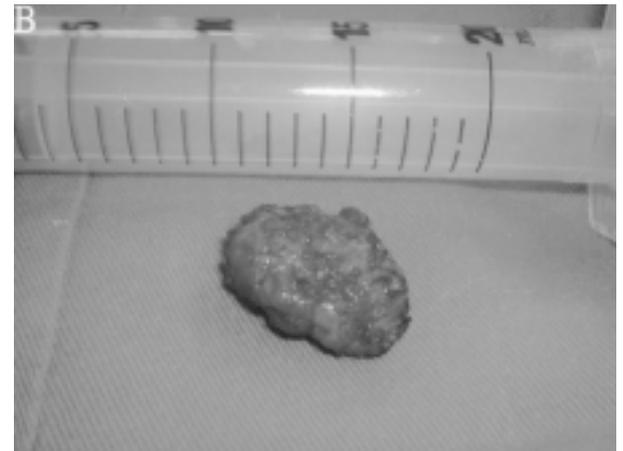


Fig. 2B Cystic mass 1.5 × 1.5-cm in size was completely excised.

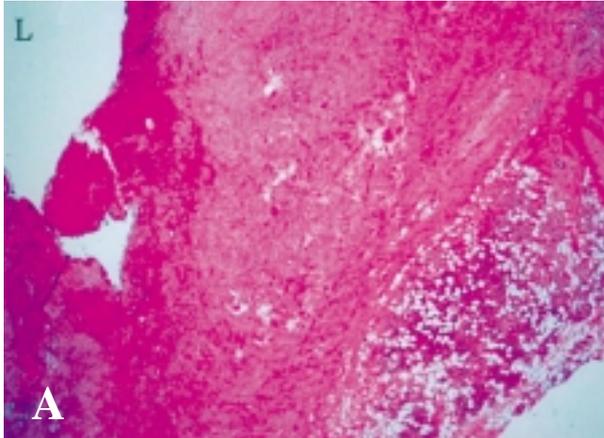


Fig. 3A Pathological examination showing the excised lesion had a cystic structure (x 100, H & E stain). L: cystic lumen;

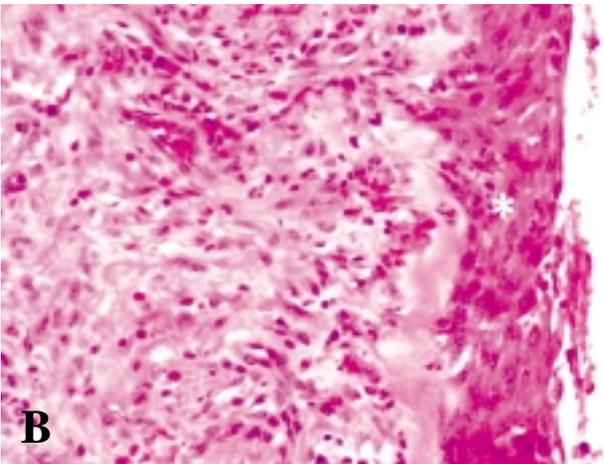


Fig. 3B Pathological examination showing the cyst wall was lined with squamous epithelium and there were mild infiltrates of lymphoplasm cells in the stroma. *: squamous epithelium (x 200, H & E stain).

DISCUSSION

Warthin's tumor is the second most common benign neoplasm of the parotid gland, accounting for 6% to 10% of all parotid tumors.⁽¹⁾ This tumor predominately occurs in older men between the fourth and seventh decades of life, with a man-to-woman ratio of 2.2:1.⁽⁵⁾ It usually presents as a slow growing mass in the parotid tail. A small group of patients, however, may present with a recent, painful and enlarged infected tumor after years of insidious behavior.⁽¹⁾ Microscopically, Warthin's tumor has a

characteristic appearance of a papillary epithelium with a lymphoid stroma projecting into cystic spaces. The epithelium consists of a double-layer of oxyphilic granular cells. The treatment of choice for Warthin's tumor is complete surgical excision, usually superficial parotidectomy.

Branchial cleft anomalies are caused by incomplete regression of the cervical sinus of His during the sixth and seventh weeks of embryologic development. They can be cysts (68%), sinuses (16%) or fistulae (16%).⁽⁶⁾ FBCCs comprise less than 1% of all branchial cleft anomalies.⁽³⁾ They are relatively rare and their embryological development remains unclear. Work classified FBCCs into two types.⁽⁴⁾ Type I, which is ectodermal in origin, is considered to be a duplication of the membranous external auditory canal. It travels parallel to the external canal towards the oropharynx. It appears as a sinus tract or a mass in the region of the pinna, external auditory canal or parotid glands. Type II is of both ectodermal and mesodermal origin and contains cartilage. It consists of a fistula running from the floor of the external auditory canal to the upper neck below the mandible. The cyst is closely associated with the parotid gland and, if infected, it usually drains near the angle of the mandible. It passes through the parotid gland and may lie either medial or lateral to the facial nerve. The relationship between type II FBCC and the facial nerve is quite variable.

The diagnosis of type II FBCCs is a clinical challenge. They can be easily misdiagnosed. Mostly, misdiagnosis is due to the rarity of type II FBCC and the unfamiliar clinical signs and symptoms. As a result, a history of multiple incision and drainage procedures for an abscess in the upper neck area is almost pathognomonic for type II FBCC.⁽⁷⁾ Occasionally, cranial nerve palsy may develop due to the intimate relationship between an FBCC and the facial nerve.⁽⁸⁾ The differential diagnosis of FBCC in children includes cystic hygroma, venous malformation, hemangioma, reactive lymphadenopathy and ectopic salivary or thyroid tissue. However, the differential diagnosis in adults should further include parotid tumor, Hodgkin's disease, lymphoma, sarcoidosis, tuberculosis and parotitis. Sichel et al.⁽⁹⁾ reported two clinical signs helpful in the diagnosis of type II FBCC. First, the location of the sinus opening in the neck, which is situated in a triangle limited by the external auditory canal above, the mental region

anteriorly and the hyoid bone inferiorly. Second, in some cases, the presence of a myringal web that runs from the floor of the external auditory canal to the umbo. A myringal web is an epidermal web that extends from the floor of the external auditory canal to the umbo of the tympanic membrane.^(10,11)

The case we presented was a case of type II FBCC masquerading as an infected parotid Warthin's tumor. The age of this patient is quite compatible with the usual manifestation of a Warthin's tumor. There was neither sinus opening nor myringal web found in this patient. Further, echo-guided aspiration cytology showed focal oncocyctic cells and the contrast enhanced CT scan showed a 2 x 2-cm, well-circumscribed cystic mass in the parotid tail. Both findings mimicked a Warthin's tumor. According to the clinical signs and symptoms, along with image and cytology studies, pre-operative diagnosis easily led to infected Warthin's tumor. However, the intra-operative finding showed that the mass was located medially to the facial nerve with a tract extending toward the external auditory canal but not communicating with it. Since the possibility of type II FBCC was highly suspected during surgery, the operative procedure changed from superficial parotidectomy to total parotidectomy, which is more challenging and often time-consuming. The incidence of facial nerve paralysis is much higher after total parotidectomy than superficial parotidectomy because stretch injury or the result of surgical interference with the vasa nervorum to the facial nerve commonly occurs after total parotidectomy.⁽¹²⁾

In diagnosing a parotid lesion, sonographic images can be valuable in distinguishing benign, cystic and malignant lesions with feasible accuracy.⁽¹³⁾ Ballo et al. reported only 81% of Warthin's tumor cytological diagnosis showed typical features; 19% of cases showed atypical images that could lead to diagnostic errors.⁽¹⁴⁾ In this case, the echo-guided aspiration cytology suggested the diagnosis of an infected parotid Warthin's tumor. CT or magnetic resonance (MR) imaging is very helpful in showing the relationship between the parotid mass and the parapharyngeal space but not in distinguishing superficial and deep lobe lesions.

Type II FBCCs are rare causes of parotid swelling and may masquerade as infected parotid Warthin's tumors, as shown by our case. It is impera-

tive that clinicians be kept aware of this rare incidence of parotid mass in differential diagnosis. They can then make an accurate pre-operative diagnosis and minimize post-operative morbidity and possible resultant recurrence.

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第二型第一對鰓裂囊腫疑似腮腺囊狀腺淋巴瘤

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腮腺腫塊的診斷，主要是依據詳細的病史和理學檢查。影像學檢查包括超音波、電腦斷層、和核磁共振，可提供重要的診斷資訊，但對於一些較少見的腮腺腫塊，幫助卻是有限。第一對鰓裂囊腫只佔所有鰓裂異常的1%，因其稀少性，再加上臨床醫師對其症狀不甚熟悉，因此常被誤診。我們報告一個第二型第一對鰓裂囊腫疑似腮腺囊狀腺淋巴瘤之病例，主要是提醒臨床醫師，稀少的第二型第一對鰓裂囊腫，可能位於腮腺的深層且以疑似腮腺囊狀腺淋巴瘤的臨床症狀來表現。我們討論第一對鰓裂囊腫之形成及臨床症狀，以期能正確的在術前診斷第二型第一對鰓裂囊腫，而避免可能的後遺症。(長庚醫誌 2006;29:435-9)

關鍵語：第一對鰓裂囊腫，腮腺腫瘤，腮腺囊狀腺淋巴瘤。

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