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Uncommon Insidious Dumbbell-shaped Double Thyroglossal Duct Cyst

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Abstract: As one of the most common congenital neck masses, thyroglossal duct cyst (TGDC) developed from the residual ductal epithelial cells in any remnants of thyroglossal duct. However, the reports of double TGDCs were rare. A 60-year-old male was referred to our department because of the presentation of an anterior neck mass. Only a hypodense oval mass inferior to hyoid bone was shown by computed tomography. During the Sistrunk operation, the dumbbell-shaped double TGDCs with the hyoid bone as the pivot were excised. No recurrence was observed. Before surgery, ultrasonography and computed tomography or magnetic resonance imaging should be conducted to verify the locations and sizes of TGDCs. During Sistrunk procedure, the rims of hyoid bone should be checked to avoid possible duct remnants.

Key Words: computed tomography, sistrunk operation, thyroglossal duct cyst, ultrasonography

Thyroglossal duct cyst (TGDC) is one of the most common congenital neck masses. Although 70% of the cases are found in children, reports suggested that there is a bimodal age distribution in the first and fifth decades.[1] However, its sexual distribution was still controversial.[1] TGDCs generally present as painless, asymptomatic soft masses in midline neck, which move with tongue protrusion and swallowing. However, the lingual subtype could cause laryngeal stridor, respiratory obstruction, and dysphagia.[2] Most TGDCs are solitary,[1] and the reports about double TGDCs were rare. Here, we presented a case of dumbbell-shaped double TGDCs with concomitant epiglottic cysts.

Though most of TGDCs locate inferior to hyoid bone, some TGDCs are found at the level of hyoid or above the hyoid and few cases are found at the base of the tongue.[2,3] Generally, the differential diagnoses of TGDCs include dermoid or epidermoid cysts, sebaceous cysts, lymphadenopathy, cystic hygromas, ectopic thyroid gland, branchial cleft cysts, hemangiomas, lipomas, bronchogenic cyst, and tuberculous lymphadenitis.[1] The close relationship between TGDC and hyoid bone is the key character for differentiation and the final differentiation relies on histopathological examination.[3] Besides, some researchers validated that subclinical persistence of TGDC could manifest it as a cyst in patients undergoing radiation therapy for head and neck cancers. In those cases, particular attentions should be paid to differentiate them from metastases.[2,4]

CLINICAL REPORT

A 60-year-old male was referred to our department because of the presentation of an anterior neck mass for 3 months. The mass could move with swallowing, which was enlarged from quail egg size to egg size. No fever, no dysphonia, no dysphagia, no hoarseness, no dyspnea, and no pharyngalgia were reported. There was no pain or swelling in neck. The patient has habits of smoking and drinking for 40 years. His neck or any other parts of body had not been subjected to radiotherapy. The remaining medical history was unremarkable.

On physical examination, a 5 × 4 cm round toughing lump was found in the midline of neck and no cervical lymphadenopathy was found. Ultrasonography revealed a cystic mass in anterior cervical region and a cyst at the base of the tongue, and there was no abnormality in thyroid screening. Routine blood and urine analysis were normal. The levels of T_3, T_4, and thyroid stimulating hormone were also normal. Rigid laryngoscopy showed cysts on lingual surface of epiglottis and multiple cysts at epiglottic valley (Fig. 1A, B). Computed tomography (CT) showed a hypodense oval mass inferior to hyoid bone, in which were internal septa. No abnormal enhancements were observed (Figs. 1C, D). The mass

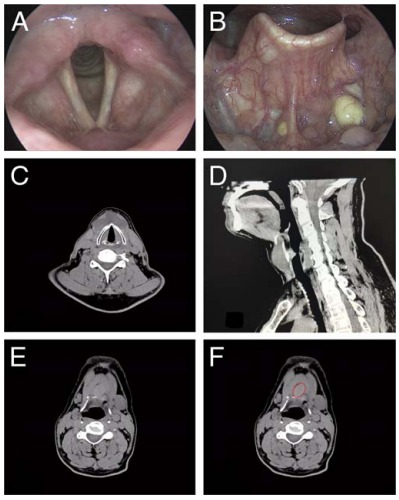

FIGURE 1. (A, B) Laryngoscopic views of the patient. Cysts were found on the lingual surface of epiglottis and at epiglottic valley. (C, D) Preoperative CT views of the cyst inferior to hyoid bone. The cyst was close to hyoid bone. (E, F) The suprahyoid isodense oval mass was identified postoperatively, which was close to hyoid bone (the mass was illustrated with red oval in F).

was cling to the hyoid bone at the midpoint. The location and size of thyroid gland were normal and no nodule was detected.

According to symptoms and physical signs, TGDC and tongue base cyst were diagnosed. Sistrunk operation was projected to remove the TGDC and explore the tongue base cyst intraoperatively. The subcutaneous fascia and muscles were separated to follow up to the mass inferior to hyoid bone. The mass is about 4 × 4 cm in size, which is located below the hyoid bone and close to the upper level of the thyroid gland. It is dark red and lobulated, with a smooth surface. Exposed the mass fully and traced it to the hyoid bone. After the mass was removed completely and the involved hyoid bone was cut off, a pale yellow cyst was seen, which was located beyond hyoid bone toward to foramen cecum. Separate the cyst till foramen cecum and resect it completely. The cyst located at tongue base was about 3.5 × 1.5 cm in size. The entire mass was dumbbell shaped with the hyoid bone as the pivot (Fig. 2). The coloboma approaching foramen cecum was sutured and reinforced without entering the pharyngeal cavity. The pathologic results validated that the cysts were lined with pseudostratified columnar epithelium and the larger cyst was multilocular, and there were inflammatory infiltration together with granulation-type tissue in the cyst wall, which verified the diagnosis of double TGDC. No evidence of malignancy was detected.

After the surgery, the CT images were carefully studied. Combined with the intraoperative presentation, one suprahyoid isodense oval mass located closely to hyoid bone was validated, which was unidentified before surgery because of its consistent dense with surrounding tissues (Figs. 1E, F).

No postoperative complication and recurrence were seen. The follow-up of patient continues.

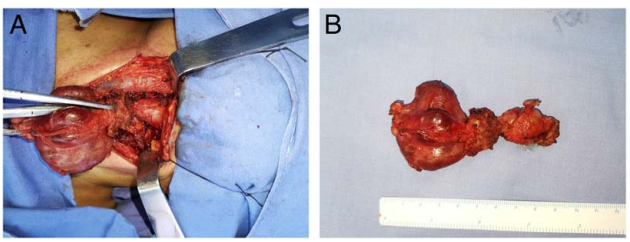

FIGURE 2. Intraoperative views and macroscopic aspect of the cysts. Both cysts were connected to hyoid bone.

DISCUSSION

As first endocrine organ developed in the embryo, thyroid anlage descends at late in the fourth week of gestation from the foramen cecum. About in the seventh week of gestation, thyroid gland arrives at its final resting point inferior to the cricoid cartilage.[1] In this process, as the trajectory for the descent of the thyroid anlage, the thyroglossal duct is developed. During seventh to tenth week of gestation, the duct generally involutes, leaving only a depression in the base of tongue. If any part of the thyroglossal duct persists, the residual ductal epithelial cells can form cysts anywhere along the passage of the thyroglossal duct.[1]

Here, we reported the fourth case in adult who was the oldest in the reported patients suffered from double TGDCs.[5–7] And this was the first reported case with concomitant epiglottic cysts. Although our results of laryngoscopy revealed several cysts at epiglottic valley, the diagnosis of epiglottic cysts were made instead of lingual TGDCs. To distinguish lingual TGDC from epiglottic cyst, direct observation of the appearance of the mass through a laryngoscope is the most common used method.[8] First, lingual TGDC usually has a thicker mucosal envelope, whereas epiglottic cysts have a thinner and more transparent envelope. Second, lingual TGDC typically locates at the midline of the tongue base inferior to foramen caecum; however, the locations of epiglottic cysts are usually lower at the level of epiglottic valley or the tongue surface of the epiglottic cartilage. Third, imaging examination can distinguish 2 kinds of disease better. The cyst next to the hyoid bone is the most diagnostic imaging feature of lingual TGDC.[8] Because no symptom was caused by those epiglottic cysts, no operations were conducted to remove them. Another issue needed to be noted is that the risk of malignant transformation of TGDCs increased with age increasing.[7] There was no malignancy in the histological examination of our case.

As summarized by Chou et al,[2] in most cases, detailed clinical history and physical examinations are adequate to make correct preoperative diagnoses. The proposes of preoperative imaging lie in confirming the diagnosis, determining the presence of the normally functioning thyroid tissue in the neck, and any possibility of malignant composition within the cyst.[5] Although CT was considered to be the gold standard imaging methods before surgical intervention by some authors,[6,9] the combined applications of ultrasonography, CT, and magnetic resonance imaging (MRI) were well suggested in literatures.[2] In our case, the dormant cyst at the base of tongue which escaped from CT screening was detected by ultrasonography before surgery. Thus, as far as we were concerned, preoperative imaging with ultrasonography and CT or MRI is of great importance to verify the locations and sizes of TGDCs, which could guarantee the identifications of insidious cysts. Besides, during the surgery, the tissues surrounding the hyoid bone should be explored meticulously to make sure that the cysts were excised exactly.

To move TGDC, Sistrunk procedure was developed, which consisted of a neck incision in which the mass and hyoid bone were exposed and subsequent removal of the mass and midportion of the hyoid bone along with a small core amount tissue of tongue base.[10] Sistrunk operation has been considered as the standard surgical management of TGDC, and the recurrence rates have been dramatically reduced.[3]

Summarizing our experience and previous reports, we suggested that preoperative imaging with ultrasonography and CT or MRI should be conducted. And after excise the TGDC and cut off the middle of hyoid bone during Sistrunk procedure, the rims of hyoid

bone should be carefully checked to detect possible duct remnants and to excise the cysts exactly, regardless of the negative results of preoperative examinations.

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Gender-Specific Differences in Chronic Subdural Hematoma

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Objective: Few studies concerning aspects of gender-specific differences in chronic subdural hematoma (CSDH). This study aimed to determine whether gender-specific differences exist in CSDH regarding clinical, radiological characteristics, and prognosis.

Methods: A total of 585 patients with CSDH were retrospectively identified. Patients were divided into 2 groups based on gender. Clinical, radiological characteristics, and prognosis were compared using Fisher's exact test or Student *t* test when applicable. The logistic regression model was used to identify independent risk factors associated with death in CSDH patients. The receiver operating characteristic curve was used to detect the sensitivity and specificity of independent risk factors.

Results: The average age of women was 71.50 ± 0.92 years, significantly older than 67.30 ± 0.60 years in men. Hypertension, diabetes mellitus, and uremia were significantly more common in women than in men. Alcohol intake was more in males than in females. CSDH patients in males manifested homogeneous iso-dense and homogeneous hyper-dense was obviously more than that in the females. Although homogeneous hypo-dense and mixed density were significantly more common in the females. The average preoperative hematoma volume of the unilateral CSDH in males was 160.85 ± 3.06 cm^3, significantly more than 139.60 ± 5.70 cm^3 in females. The mortality of females was 7.4%, higher than 1.7% in males ($P = 0.004$). Female, age, uremia, and recurrence were independent risk factors for death in CSDH patients.

Conclusions: Gender-specific differences do exist in CSDH. Female, age, uremia, and recurrence were independent risk factors for death in CSDH patients.

Key Words: chronic subdural hematoma, glasgow coma scale, glasgow outcome, gender, mortality

Chronic subdural haematoma (CSDH) is a common disorder with increasing prevalence among seniors. The annual incidence of CSDH is 16.3/100,000 people in patients younger than 65 years, and up to 58.1/100,000 people in patients aged 65 years and older.[1]

A well-known characteristic of CSDH is its male predominance. The male/female ratio has been reported to range from 1.7:1 to 4.7:1.[2–5] Biological and behavioral differences between males and females affect the epidemiology, pathophysiology, clinical manifestations, and prognosis of many diseases. Understanding the gender-specific differences in diseases helps individualized treatment and postoperative management of patients.[6–9]

However, few studies concern the aspects of gender-specific differences in CSDH patients. In the present study, we aimed to determine whether gender-specific differences exist in CSDH patients regarding clinical, radiological characteristics, and prognosis.

PATIENTS AND METHODS

Exclusion Criteria

In this retrospective study, we included 585 consecutive CSDH patients treated in our Department between October 2012 and November 2019. Exclusion criteria as the following: (1) patients who are younger than 18 years old; (2) CSDH caused by arachnoid cyst; (3) CSDH caused by cerebrovascular diseases, such as aneurysms or vascular malformations; (4) underwent other intracranial operation

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We obtained ethical approval to carry out this study from the Institutional Review Board (IRB) of Yijishan Hospital of Wannan Medical College, Wuhu, China of Tianjin Medical University General Hospital, Tianjin, China. All patients provided written informed consent for this study, which was carried out in accordance with the Helsinki Declaration. All individual information was kept strictly confidential and anonymous in the manuscript.

The authors report no conflicts of interest.

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