

Original Article

Recurrent acute suppurative thyroiditis secondary to pyriform sinus fistula: report of two cases and literature review

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Abstract: Acute suppurative thyroiditis (AST) is a rare infectious condition as we know the thyroid gland is remarkably resistant to infection. Pyriform sinus fistula is an extremely rare branchial apparatus malformation but considered the leading cause of AST. Here, we report two cases of AST secondary to pyriform sinus fistula. A 14-year-old girl presented with a swelling in the anterior neck with tenderness and fever for 15 days. Similar symptoms appeared 2 and 5 months ago. AST was confirmed diagnostically by blood tests and imaging. Her condition was significantly improved by sensitive antibiotics along with catheter drainage. However, dilute barium swallow examination indicated a possible fistula connected to the pyriform sinus. Following a successful surgery to the fistula, she recovered uneventfully within 25 days. On one year follow-up, no fistula was detected. The second case is a 30-year-old man who complained of fever for 20 days with tenderness and swelling in the neck anteriorly for 15 days. The patient experienced similar manifestations 2 and 10 years ago, of which the previous diagnosis and treatment was unknown. Blood tests and imaging examinations were obtained, followed by the diagnosis of AST. Fever was relieved and tender swelling in the neck was reduced significantly with sensitive antibiotics and drainage of pus. Discovered by barium swallow radiology, the pyriform sinus fistula was successfully removed by radical resection without recurrence on one year follow-up.

Keywords: Acute suppurative thyroiditis, pyriform sinus fistula, thyroid abscess

Introduction

Acute suppurative thyroiditis (AST) is a rare infectious condition as the thyroid gland is remarkably resistant to infection because of its high iodine content, rich blood supply and lymphatic drainage and protective fibrous capsule of the gland [1, 2]. Furthermore, any thyroid suppurative inflammation, particularly recurrent infections in normal immunity population should raise a suspicion for the presence of predisposing conditions. Most commonly, the third or fourth pharyngeal pouch forms pyriform sinus fistula, resulting in a tract between the thyroid and the throat, a common route of infection in AST. The diagnosis of AST mainly depends on medical imaging such as computed tomography (CT) and endoscopy. Treatment with antibiotics and/or incision and drainage is indicated during acute exacerbations. Perma-

ent cure of pyriform sinus fistula can only be achieved by complete fistulectomy or chemo-cauterization [3].

Case 1

A previously healthy 14-year-old girl complained of anterior neck tender swelling and fever for 15 days. The highest temperature recorded was 39.5°C. Similar symptoms were shown 2 and 5 months ago. She was diagnosed with acute anterior neck suppurative disease in primary hospital, and treated only with antibiotics. Physical examination revealed a 5 cm×4 cm sized mass in the anterior neck with tenderness as well as redness and warmth in the overlying skin. Blood tests showed white blood cell $18.26 \times 10^9/L$ with 92.3% neutrophils, high sensitive C-reactive protein (hsCRP) 51.6 mg/L, erythrocyte sedimentation rate (ESR) 87 mm/h,

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Figure 1. Barium swallow examinations revealed an approximately 3 cm long fine barium shadow.



Figure 2. Barium swallow examinations revealed an approximately 2.5 cm long fine barium shadow.

and normal fT3, fT4 and TSH. Thyroid ^{131}I scan showed a diffusive decrease in the enlarged left lobe of thyroid. Ultrasound scan and CT revealed a 4 cm×3 cm sized abscess involving the left lobe of the thyroid gland and multiple reactive cervical lymphadenopathy, but no definite evidence of fistula connected to the pyriform sinus. Ultrasound guided fine needle aspiration was performed on the 2th day of admis-

sion and about 10 mL yellow pus was collected. On 7th day of admission, hemolytic streptococcus was isolated from pus and susceptibility testing showed it was sensitive to cefoperazone-sulbactam, so clindamycin was replaced by cefoperazone-sulbactam (2.0 g, bid). Her symptoms gradually disappeared along with WBC, hsCRP and ESR decreased gradually. Dilute barium swallow test was taken on 14th day of admission when the symptoms were significantly reduced. An approximately 3 cm long fine tract was discovered extending left laterally to the bottom of the pyriform sinus, which was demonstrated below (**Figure 1**). On 18th day of admission, this patient was transferred to department of otorhinolaryngology-head and neck surgery and underwent the surgery of radical resection of fistula. After surgery, dilute barium swallow test showed no tract image. He was discharged at the 25th day with an uneventful postoperative follow-up after 1 year, and there were no any complications.

Case 2

A previously healthy 30-year-old man complained of fever for 20 days and a tender swelling in the anterior neck for 15 days. His highest documented temperature was 39.2°C. Symptoms were noticed 2 and 10 years ago and the previous diagnosis and treatment were unknown. On admission, the vital signs were temperature 39.1°C, pulse rate 100/min, and BP 120/70 mmHg. Physical examination revealed a 6 cm×4 cm sized anterior neck mass with tenderness. Laboratory tests showed white blood cell count $22.36 \times 10^9/\text{L}$ with 89.2% neutrophils, hsCRP 68.3 mg/L, ESR 112 mm/h and serum fT3, fT4 and TSH level normal. A diffusive decrease of ^{131}I uptake was indicated in the enlarged left lobe of thyroid. A 5 cm×3 cm sized abscess involving the left lobe of the thyroid gland was revealed by both ultrasonography and CT. However, there was no definite evidence of a fistula communication to the pyriform sinus. Ultrasound-guided fine needle aspiration was performed on the 3th day of hospitalization, and pus was drained continually through the incision site until the fever and neck swelling subsided without further suppurative production. About 10 mL yellow pus was collected to be given bacterial culture and antibiotic sensitivity tests. On 9th day, escherichia coli was isolated from pus, which was sensitive

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Table 1. Summary of previous reported similar cases of

Authors	Number of cases	Sex (male/female)	Childhood/Adult	Average number of episode	Detection of PSF by barium swallow	Prognosis (number of cured)
Parida PK et al. [4]	17	5/12	17/0	3	15	16
Cieszynski L et al. [5]	1	0/1	0/1	5	1	1
Yolmo D et al. [6]	15	6/9	15/0	Not reported	12	15
Schneider U et al. [7]	1	0/1	1/0	4	1	1
Seo JH et al. [1]	1	0/1	1/0	Not reported	1	1

to levofloxacin. With antibiotic replacement of levofloxacin (0.5 g, qd), the above symptoms gradually disappeared, so as WBC, hsCRP and ESR gradually returned to normal ranges. On 20th day, when his fever and anterior neck tender swelling disappeared, dilute barium swallow test was performed, which revealed the fistula connected to the pyriform sinus (**Figure 2**). On the 22th day, he was transferred to department of otorhinolaryngology-head and neck surgery and underwent complete fistulectomy. He was discharged at the 30th day satisfactorily, and there were no any complications on 1 year follow-up.

Literature review

To identify previously reported cases of recurrent acute suppurative thyroiditis secondary to pyriform sinus fistula, we searched the PubMed database using the search words "Recurrent acute suppurative thyroiditis" and "pyriform sinus fistula". In addition, the references cited in previously published reports were also reviewed. 35 cases were identified and results are summarized in **Table 1**. 34 patients (97%) were childhood and only 1 patient (3%) was adult. All patients presented with fever and left thyroid lobe abscess. US and CT of the neck showed the abscess involving the left lobe of thyroid gland in all patients. The pyriform sinus fistula tract was demonstrated with barium swallow examinations in 30 out of 35 (85.7%) patients. Complete excision of pyriform sinus fistula is an effective method to prevent recurrence of acute suppurative thyroiditis in most patients.

Discussion

AST is a rare clinical condition as the thyroid gland is remarkably resistant to infections due to its rich blood and lymphatic supply, high iodine content and protective fibrous capsule.

Pyriform sinus fistula, being an extraordinarily unusual appearance of branchial apparatus malformation, is most responsible for the occurrence of AST. So thyroid suppurative infection, particularly recurrent infections in normal immunity population, should raise a suspicion for the presence of pyriform sinus fistula. Third and fourth branchial remnants can present at any age from infancy to adulthood, as a result AST due to pyriform sinus fistula may occur at any age, but most common in childhood [8].

Clinical features of AST include thyroid abscesses, recurrent and non-healing sinus in the lateral aspect of the neck, fever, cough, hoarseness, dysphagia, sore throat [9]. Both thyroid gland lobes can be affected but the left lobe is more frequently involved and this is possibly related to asymmetry of the fourth branchial arch to form the aortic arch and innominate artery [10]. Leukocytosis and increased ESR and CRP are common. Thyroid function tests are usually normal, although thyrotoxicosis may occur in 5% and transient or permanent hypothyroidism can occur in about 2%-3% of patients as a result of the disease or treatment [11, 12]. Clinical diagnosis demands a high index of suspicion as these anomalies are rare and present with distressing recurrences if not excised completely. Ultrasonography often serves as the initial imaging modality for the assessment of painful neck masses, usually showing heterogeneous echogenicity of the thyroid gland with irregular margin and inflammatory changes. CT scan is also a useful modality for detecting loss of the normal high density of the thyroid gland, abscess formation and air density of a sinus or fistula. If an abscess or inflammation in the thyroid lobe is detected, further imaging is required to exclude an underlying pyriform sinus fistula especially if it is recurrent and left sided [13]. Usually, pyriform sinus fistula was not easily identified by CT or ultrasonography. Also based on the fact that the pres-

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ence of pyriform sinus fistula in our two cases was detected by dilute barium esophagram instead of CT and ultrasonography, we think dilute barium swallow examination is perhaps more sensitive to detect the pyriform sinus fistula than other medical imaging. But it must be noted that dilute barium rather than common barium was used while barium swallow being performed, because dilute barium was easier to pass through the slender sinus. In addition, it is very important to choose the right time of the test. Usually the best is after the inflammation subsided, because in the acute inflammatory phase the fistula is likely to be closed and difficult to be identified due to edema or other reasons. Laryngoscopy or hypopharyngoscopy can also detect underlying anatomical defects, through which, however, it is difficult to find the fistula opening in the acute inflammatory period or when the fistula is very slender. Laryngoscopy was performed in our two cases after the inflammation subsided, but, not surprisingly, only local mild edema was discovered in laryngopharynx with no identification of the fistula opening, as expected in a narrow passage of the fistulae in these two cases.

Therapy with empiric antibiotics includes penicillinase-resistant penicillin or combination of a penicillin and a β -lactamase inhibitor should be used in early phase. Penicillin, clindamycin, combination of macrolide and metronidazole is sometimes recommended. When thyroid abscess is formed, Ultrasound-guided fine needle aspiration and catheter drainage should be performed. Some of the patients may receive abscess incision and drainage. Pus must be collected for bacterial culture and drug sensitivity test. Following the test results, antibiotics should be adjusted accordingly. To prevent the reoccurrence of acute suppurative thyroiditis due to the presence of pyriform sinus fistula, radical resection of fistula is considered a definitive treatment for the disease [14].

In conclusion, AST is a rare lesion predominantly involving the left lobe of the thyroid and almost always secondary to pyriform sinus fistula. Its main clinical symptoms are fever and anterior neck swelling. Although ultrasonography and CT often serve as important methods for the diagnosis of AST, dilute barium swallow is more sensitive to detect pyriform sinus fistula. Surgery of radical resection of fistula is an approach to cure this disease.

Disclosure of conflict of interest

None.

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