

Recurrent Acute Suppurative Thyroiditis in A Patient without Pyriform Sinus Fistula : A Case Report

Hsiao-Lien Chen¹, and Pai-Lien Chen²

*¹Division of Endocrinology and Metabolism, Department of Medicine,
Lo-Tung Poh-Ai Hospital, Ilan County, Taiwan*

*²Division of Endocrinology and Metabolism, Department of Medicine,
Lo-Tung Saint Mary's Hospital, Ilan County, Taiwan*

Abstract

Acute suppurative thyroiditis is a rare disorder that causes severe pain, tenderness, redness and fluctuation in the region of the thyroid gland. A 52-year-old healthy woman had acute onsets of left neck painful swelling on two occasions, three years ago and one year ago, respectively. A sonography examination revealed a hypoechoic area on the left lobe of thyroid, about 2.28 x 2.1cm in size; a fine-needle aspiration biopsy showed neutrophil infiltration. Appropriate antibiotic treatment and incision and drainage of mass relieved the symptoms of the infection. The patient had no history of diabetes mellitus and no sign of pyriform sinus fistula on barium swallow study; it is unusual to develop recurrent suppurative thyroiditis in such a patient. (J Intern Med Taiwan 2007; 18: 134-139)

Key Words : Acute suppurative thyroiditis; Pyriform sinus fistula

Introduction

Acute suppurative thyroiditis (AST) is an uncommon infectious disease that affects mainly children and young adults¹. The rarity of AST is a result of the resistance of the thyroid gland to local infection. The resistance of the gland to disease results

from its complete encapsulation, general vascular supply, lymphatic drainage, and local presence of iodine, which inhibits bacterial proliferation². Anatomical abnormalities, previous diseases of the gland, sepsis or immunologic disturbances may reduce resistance and increase the number of infection events²⁻⁵.

Correspondence and requests for reprints : Dr. Hsiao-Lien Chen

Address : Division of Endocrinology and Metabolism, Department of Internal Medicine, Lo-tung Poh-Ai Hospital, Ilan County, Taiwan 83 Nan Chang Street, Lotung, Ilan, 265 Taiwan, R.O.C.

Case Report

A 52-year-old healthy woman who had no systemic disease and diabetic, presented with fever and painful swelling in the anterior side of the neck for one day. The neck pain was associated with fever, sore throat and dysphagia; the pain radiated to the left ear.

Three years ago, the patient had suffered an AST attack, with a $3.5 \times 3\text{cm}$ tender fluctuant mass at the left thyroid gland. The patient was admitted to our hospital following ten days illness characterized by fever, sore throat, neck pain despite treated with antibiotic and non-steroidal anti-inflammatory drugs at primary care physician. Ultrasonography and a neck

computed tomography (CT) scan revealed a lobulated hypodense lesion at the left side of thyroid gland (Figure 1, 2). A fine-needle aspiration biopsy obtained a small amount of thick fluid. A cytology of the thyroid aspirate revealed numerous neutrophils. Bacterial culture of aspirate was done but no bacterial growth. The patient was treated with amoxicillin/clavulanic acid for seven days, and the mass was incised and drained on the second day following admission. The blood culture showed no bacterial growth. The patient was discharged after full recovery and the outpatients department has conducted, but no subsequent follow-up.

The physical examination during the more recent event revealed that the patient's body temperature was 37.6°C , her pulse rate was 80 beats per minute, and that there was a tender fluctuant mass measuring up to 2.5cm in diameter, with an erythematous skin lesion in the left neck. Lymphadenopathy was not present. Laboratory tests showed a leukocyte count of 13000/uL, an erythrocyte sedimentation rate (ESR) of 34mm per hour and 64mm per two hours, thyroid functional tests within the normal range and the absence of thyroid antibodies. Ultrasonography of the thyroid revealed two hypoechoic masses at the left lobe of the thyroid, approximately $2.28 \times 2.1\text{cm}$ in size and 1.57cm in diameter (Figure 3). Fine-needle aspiration biopsy obtained approximately



Fig.1.Sonogram on first attack showed hypoechoic lobulated mass at left lobe of thyroid



Fig.2.Neck CT on first attack showed lobulated hypodense lesion at left side of thyroid



Fig.3.Sonogram on second attack showed hypoechoic lobulated mass at left lobe of thyroid

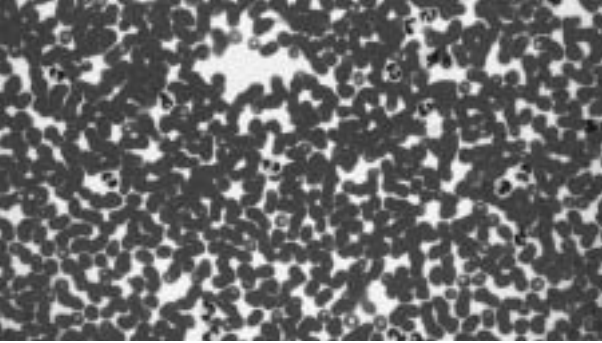


Fig.4. Fine-needle aspiration biopsy of the thyroid showed many neutrophils (400X), Papanicolaou stain



Fig.5. ^{99m}Tc scan revealed diminished uptake at left lobe, particularly on left upper lobe

0.1cc of thick, yellowish fluid and the smear revealed numerous neutrophils and scanty follicular cells (Figure 4). No culture of the fluid was done. Thyroid scan revealed diminished uptake of $\{^{99m}\text{Tc}\}$ throughout the left lobe, with normal uptake in right lobe (Figure 5). A neck CT scan showed a lobulated hypodense lesion with rim enhancement on the left side of the thyroid gland; an extension from the level of the epiglottis to the level of the thoracic inlet was compressing the airway (Figure 6). The blood culture also showed no growth on this occasion. The patient was treated with systemic antibiotic amoxicillin/clavulanic acid for six days, and the incision and drainage of the left thyroid abscess were arranged on the second day following admission. A barium swallow study was performed one month and three

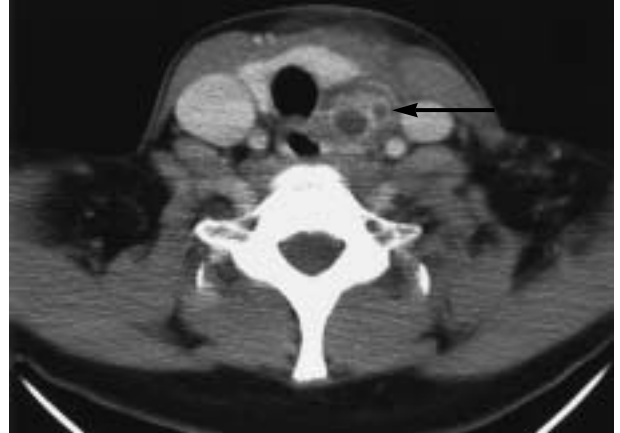


Fig.6. Neck CT scan on second attack showed hypodense lesion with abnormal rim enhancement at left side



Fig.7. Barium swallow study showed no signs of pyriform sinus fistula

months later; there was no signs of pyriform sinus fistula (Figure.7).

Discussion

AST in the thyroid frequently occurs as an ex-

tension from the adjacent structures, by direct trauma or hematogenous seeding from a distant focus. The predisposing factors to AST in adults include immunocompromised, structural abnormalities of the thyroid gland, and infection of the skin and lower respiratory tract^{1,17}. Occasionally, the source of the infection is a persistent thyroglossal duct or pyriform sinus fistula²⁻⁴.

The literature review revealed that the recurrent AST due to the presence of persistent pyriform sinus thyroid fistula 80% occurred at age 0-10 years (mean age 7.6 years)^{5,11-15,17}; 30% occurred at age 0-2 years^{5,6,11,12,14,15} and 8% in adulthood⁷⁻¹¹.

Ninety-two percent of cases involved the left thyroid lobe⁵⁻¹¹, 6% the right lobe¹¹⁻¹³, and 2% the bilateral lobes^{14,15}. Over 90% of the patients with AST presented with thyroidal pain, tenderness, fever and local compression, leading to dysphagia and dysphonia^{18,20}.

Normal thyroid function tests are seen in over two thirds of patients with acute suppurative thyroiditis^{1,19,20} although thyrotoxicosis and hypothyroidism have been reported^{20,21,22}. Leukocytosis and the elevation of both ESR and C-reactive protein level were observed in both subacute and infectious thyroiditis. The majority of studies showed that the thyroid function status was euthyroid in AST, with only a few case reports showing temporary hyperthyroidism in the acute stage^{1,3,16}.

Fine-needle aspiration biopsy is the best single laboratory test for the evaluation of infectious thyroiditis, and is used in the diagnosis of most cases^{19,23,24}, especially when the tenderness is limited to a solitary nodule or localized area and when the disease of subacute thyroiditis has been ruled out. A culture of aspirate should be performed to guide antimicrobial therapy.

Aggressive malignant thyroid tumors may mimic the clinical symptoms and signs of AST in the early stage of the disease. Risk factors that differentiate malignant thyroid tumor from AST include older age,

the presence of dysphonia, the involvement of the right lobe, a larger thyroid mass, and the presence of anemia and sterile pus culture¹⁶.

The diagnosis of a persistent pyriform sinus-thyroid fistula requires an index of suspicion. The presence of the persistence of pyriform sinus fistula should be considered when a recurrent left neck abscess or acute thyroiditis fails to respond to the appropriate antibiotic and surgical therapy. Barium swallow is considered to be an effective means of demonstrating the presence of an anomalous tract, with a sensitivity of 80%^{5,7-10,12-13}. False negative results were mostly due to edema around the orifice of the tract^{5,9,11-14}, which occur during acute infection and may prevent the contrast medium from entering the sinus tract²². Thus, the optimal time for this study is during the quiescent stage following antibiotic therapy.

In the treatment of AST, systemic antibiotic therapy should be applied as soon as possible and surgical drainage should be considered when fluctuation is observed. In the cases of recurrent AST with persistent pyriform sinus fistula, cure is achieved only by a complete resection of the sinus tract, including the portion of the thyroid at which the sinus tract terminates^{5,7,9,25}. Incision, drainage and antibiotic treatments may be used as temporizing measures prior to the definite surgery.

In our case, recurrent suppurative thyroiditis occurred at the left lobe on two occasions in two years; the fistula was highly suspicious. On this occasion, we arranged barium swallow at one month and three months after the second attack of suppurative thyroiditis; no sign of pyriform sinus fistula was found.

In conclusion, AST is an uncommon disease, particularly if recurrent. It is more common in childhood and associated with pyriform sinus-thyroid fistula. We report a case of recurrent AST that occurred in healthy adult patient with no evidence of pyriform sinus fistula. Treatment with antibiotics, incision and drainage is the definitive treatment. Careful follow-

up with thyroid sonography and the constant observation of inflammatory signs and symptoms are required for this type of patient.

References

1. Berger SA, Zonszein J, Villamena P, et al. Infectious diseases of the thyroid gland. *Rev Infect Dis* 1983; 5: 108-22.
2. Premawardhana LD, Vora JP, Scanlon MF. Suppurative thyroiditis with oesophageal carcinoma. *Postgrad. Med J* 1992; 68: 592-3.
3. Miyauchi A, Matsuzuka F, Kuma K, et al. Piriform sinus fistula: an underlying abnormality common in patients with acute suppurative thyroiditis. *World J Surg* 1990; 14: 400-5.
4. Chiovato L, Canale G, Maccherini D, et al. Salmonella Brandenburg: a novel cause of acute suppurative thyroiditis. *Acta Endocrinol* 1993; 128: 439-42.
5. Cases JA, Wenig BM, Silver CE, et al. Recurrent acute suppurative thyroiditis in an adult due to a fourth branchial pouch fistula. *J Clin Endocrinol Metab* 2000; 85: 953-6.
6. Raven RW. Pouches of the pharynx and esophagus with special reference to the embryological and morphological aspects. *Br J Surg* 1933; 21: 235-6.
7. Takai S, Matsuzuka F, Miyauchi A, et al. Internal fistula as a route of infection in acute suppurative thyroiditis. *Lancet* 1979; 1: 751-2.
8. Miyauchi A, Matsuzuka F, Takai S, et al. Piriform sinus fistula. *Arch Surg* 1981; 116: 66-9.
9. Nonomura N, Ikarashi F, Fujisaki T, et al. Surgical approach to pyriform sinus fistula. *Am J Otolaryngol* 1993; 14: 111-5.
10. Yamashita J, Ogawa M, Yamashita S, et al. Acute suppurative thyroiditis in an asymptomatic woman: an atypical presentation simulating thyroid carcinoma. *Clin Endocrinol* 1994; 40: 145-50.
11. Bar-Ziv J, Slasky BS, Sichel JY, et al. Branchial pouch sinus tract from piriform fossa causing acute suppurative thyroiditis, neck abscess, or both: CT appearance and the use of air as a contrast agent. *Am J Roentgenol* 1996; 167: 1569-72.
12. Ahuja AT, Griffiths JF, Roebuck DJ, et al. The role of ultrasound and oesophagography in the management of acute suppurative thyroiditis in children associated with congenital pyriform fossa sinus. *Clin Radiol* 1998; 53: 209-11.
13. Katz R, Bar-Ziv J, Preminger-Shapiro R, et al. Pyogenic thyroiditis due to branchial pouch sinus. *Isr J Med Sci* 1989; 25: 641-4.
14. DeLozier HL, Sofferman RA. Pyriform sinus fistula. an unusual cause of recurrent retropharyngeal abscess and cellulites. *Ann Otol Rhinol Laryngol* 1986; 95: 377-82.
15. Shaw A. Acute suppurative thyroiditis. *Am J Dis Child* 1979; 133: 757.
16. Lin KD, Lin JD, Huang MJ, et al. Acute suppurative thyroiditis and aggressive malignant thyroid tumors: differences in clinical presentation. *J Surg Oncol* 1998; 67: 28-32.
17. Chang P, Tsai WY, Lee PI, et al. Clinical characteristics and management of acute suppurative thyroiditis in children. *J Formos Med Assoc* 2002; 101: 468-71.
18. Jeng LB, Lin JD, Chen MF. Acute suppurative thyroiditis: a ten-year review in a Taiwanese hospital. *Scand J Infect Dis* 1994; 26: 297-300.
19. Rich EJ, Mendelman PM. Acute suppurative thyroiditis in pediatric patients. *Pediatr Infect Dis J* 1987; 6: 936-40.
20. Szabo SM, Allen DB. Thyroiditis: Differentiation of acute suppurative and subacute: case report and review of the literature. *Clin Pediatr (Phila)* 1989; 28: 171-4.
21. Walsh CH, Dunne C. Hyperthyroidism associated with acute suppurative thyroiditis. *Ir J Med Sci* 1992; 161: 137.
22. Fukata S, Miyauchi A, Kuma K, et al. Acute suppurative thyroiditis caused by an infected piriform sinus fistula with thyrotoxicosis. *Thyroid* 2002; 12: 175-8.
23. Lin JD, Huang BY, Huang HS, et al. Ultrasonography and fine needle aspiration cytology of acute suppurative thyroiditis: *Chang Gung Med J* 1993; 16: 93-9.
24. Singh SK, Agrawal JK, Kumar M, et al. Fine needle aspiration cytology in the management of acute suppurative thyroiditis. *Ear Nose Throat J* 1994; 73: 415-7.
25. Karlan M S, Michel SL, Snyder WH Jr. Branchiogenic cysts: congenital or acquired. *Am J Surg* 1965; 110: 615-9.

急性復發化膿性甲狀腺炎在無 pyriform sinus 瘻管之病患：一病例報告

陳曉蓮¹ 陳白蓮²

¹ 羅東博愛醫院 內科部內分泌暨新陳代謝科

² 羅東聖母醫院 內科部內分泌暨新陳代謝科

摘 要

急性化膿性甲狀腺炎是一種少見的疾病，臨床表徵為甲狀腺腫大、皮膚表面有發紅、壓痛、發燒…等。我們報告一位五十二歲的女性，因左側甲狀腺腫大且疼痛、發燒住院治療。甲狀腺超音波發現左側甲狀腺有2.28×2.1公分大腫塊、用細針抽取內容物發現許多白血球。在急性化膿性甲狀腺炎之診斷下，給予抗生素治療及切開腫塊後改善病患症狀。患者痊癒三個月後接受barium swallow study的檢查，未發現瘻管。病患參年前此症狀，經過藥物治療及手術引流膿瘍後便全癒。此病患為中年女性無糖尿病或免疫機能不足病史，但兩年內反覆發生化膿性甲狀腺炎並未發現pyriform sinus瘻管是個少見案例。