



Recurrent acute suppurative thyroiditis due to pyriform sinus fistula in an adult — case report

Nawracające ropne zapalenie tarczycy a przetoka zachyłka gruszkowatego — opis przypadku

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Abstract

We present a case study of acute suppurative thyroiditis (AST) with frequent recurrence due to anatomical malformation described as pyriform sinus fistula (PSF). Difficulty in diagnosis and treatment may be explained by the rarity of the disease in adult patients. AST had been observed in our patient five times before a radical surgery treatment was performed. The most typical symptoms and signs in our patient include: fever, difficulty in swallowing and tender tumour in the left side of the neck. Computed tomography (CT), barium swallow study and endoscopic examination gave us an opportunity to confirm the presence of an anatomical abnormality. After fistulectomy, we have not observed AST up to the date of this publication. (*Endokrynol Pol* 2013; 64 (3): 234–236)

Key words: suppurative thyroiditis, pyriform sinus fistula, adults

Streszczenie

W pracy przedstawiono opis przypadku nawracającego bakteryjnego zapalenia tarczycy w przebiegu nieprawidłowości anatomicznej określanej mianem przetoki zachyłka gruszkowatego części kraniowej gardła. Trudności w diagnostyce i leczeniu wynikają z rzadkiego występowania omawianego problemu u ludzi dorosłych. Bakteryjne zapalenie tarczycy stwierdzono u pacjentki 5-krotnie zanim podjęto leczenie operacyjne. Objawy kliniczne zapalenia tarczycy obejmowały: gorączkę, trudności w polykaniu, bolesny guz po lewej stronie szyi. Badania obrazowe: tomokomputerowe szyi, kontrastowe górnego odcinka przewodu pokarmowego z użyciem barytu oraz endoskopia, potwierdziły obecność omawianej malformacji anatomicznej. Chirurgiczne usunięcie przetoki zapobiegło kolejnym nawrotom bakteryjnego zapalenia tarczycy. (*Endokrynol Pol* 2013; 64 (3): 234–236)

Słowa kluczowe: ropne zapalenie tarczycy, przetoka zachyłka gruszkowatego, dorośli

Introduction

Acute suppurative thyroiditis (AST) due to pyriform sinus fistula (PSF) is a very rare disease in adults. There have been only a few cases reported in the English language literature, mostly in otolaryngology, surgery and paediatric journals. A possible difficulty in diagnosis and treatment may also be of interest to endocrinology clinicians.

We present a case report where there was a long gap between the first treatment and the most recent procedure. The clinical symptoms, diagnosis and treatments are analysed here.

Case report

A 26 year-old woman was admitted, for the first time, to the Outdoor Department of Surgery in April 2007.

She presented with symptoms of abscess formation in the left frontal neck area. Treatment with antibiotics, nonsteroidal anti-inflammatory drugs, incision and drainage of abscess were performed. Positive cultures for multiple pathogens were obtained: *Streptococcus viridans*, *Fusobacterium nucleatum*, *Peptostreptococcus asaccharolyticus* and *Prevotella melaninogenica*. No additional specialist diagnostic was undertaken.

In February 2009, she was referred to our Department because of recurrence of the disease. She had fever, a shapeless mass in the neck, and difficulty in swallowing. On examination, we discovered a tender tumour in the left side of the neck; the overlying skin was erythematous and no lymphadenopathy was observed. Laboratory investigations revealed positive C reactive protein, an increased erythrocyte sedimentation rate, and mild suppression of thyrotropin (TSH) with serum free triiodothyronine (fT3)



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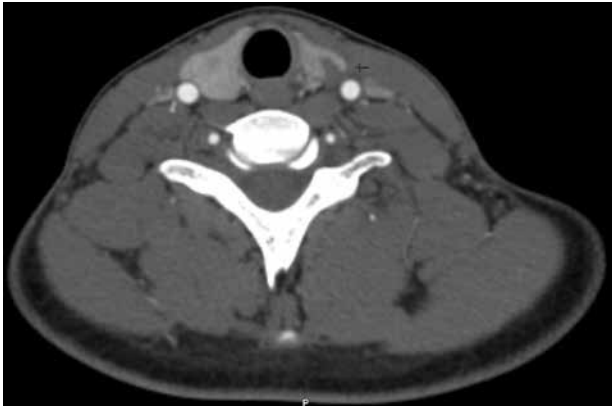


Figure 1. Neck CT showing inflammation of the left lobe of the thyroid gland

Rycina 1. TK szyi wykazujące zapalenie w lewym płacie tarczycy

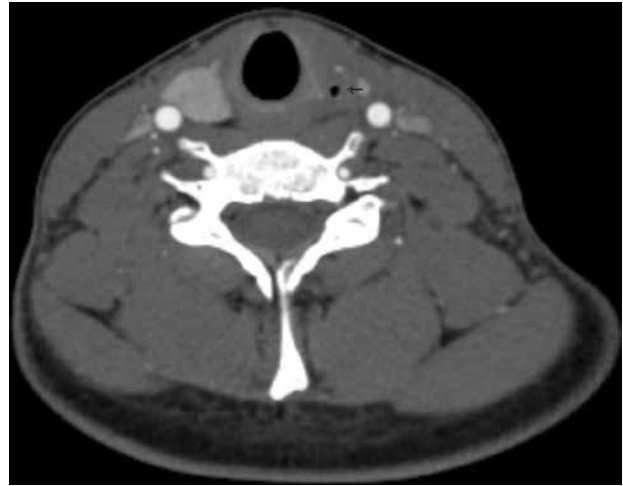


Figure 2. Neck CT showing possible pyriform sinus fistula on the left side

Rycina 2. TK szyi uwidaczniające prawdopodobny przebieg przetoki zachyłka gruszkowatego po stronie lewej



Figure 3. Barium meal study showing fistula of the left pyriform sinus

Rycina 3. Badanie z użyciem barytu uwidaczniające przetokę zachyłka gruszkowatego po stronie lewej

elevations. Computed tomography (CT) with longitudinal section reconstruction showed an enhanced lesion in the superior area suggesting inflammatory state (Fig. 1 and 2).

A diagnosis of acute suppurative thyroiditis (AST) due to possible pyriform sinus fistula (PSF) was made. The patient responded well on intravenous administration of antibiotics. To prevent recurrence of the inflammation, we proposed fistulectomy, which she declined.

In November 2010, the patient reported with next AST. Ultrasonography revealed a liquid cystern corresponding most likely with an abscess formation.

Fine-needle aspiration biopsy enabled us to evacuate approximately 1.5 cm³ of liquid content of the abscess. Both *Streptococcus beta-haemolyticus* Group F and *Streptococcus alpha-haemolyticus* pathogens were identified. To confirm PSF, barium swallow study and endoscopic examination were performed one and two months after the acute inflammatory phase respectively. Barium meal study demonstrated fistula originating from the apex of left pyriform sinus (6 mm in diameter), connected with a grindstone-shaped cistern (12 x 2.5 mm in dimension) (Fig. 3).

Direct hypopharyngoscopy under general anaesthesia indicated an internal orifice leading to the fistula.

In August 2011, she was readmitted to our Department for the final evaluation because of a further AST recurrence. Finally the patient gave her consent to fistulectomy. Treatment with co-trimoxazole for AST prophylactic before the planned surgery resulted in agranulocytosis. Discontinuation of antibiotic treatment provoked subsequent infective thyroiditis, finally medicated with amoxicillin.

In November 2011, fistulectomy and partial thyroidectomy were performed. No pathologic detection of fistula was made; microscopic sections included thyroid structures with granulation and fibrous tissue. Since that time, we have not observed AST.

Discussion

AST is a rare disease [1]. Up to 2008, only 28 adult patients with AST due to a PSF had been described in the English language literature [2, 3]. Most of the reported cases were related to paediatric patients, whereas only a few cases concerned adult patients [4–6]. Thyroid

gland resistance to local infection might be explained by complete encapsulation, rich blood supply, effective lymphatic drainage and the inhibitory action of high iodine content. Anatomical abnormalities such as persistent thyroglossal duct and PSF are direct communication resulting in infectious thyroiditis [2, 7, 8]. PSF complicated with AST usually affects 80% of patients during their first decade of life, but only 8% in adulthood [2, 4, 5, 16].

At present, the embryological development of PSF seems to be controversial. Traditional theory of fistula formation considers 3rd and 4th branchial arch origin [5, 8–10, 22]. It is explained by histopathological presence of thymic tissue close to fistula in some cases and anatomical location in the course of laryngeal nerves embryonic tract. On the other hand, the spatial maldistribution of C cells observed in the thyroid gland suggests that a remnant of the ultimobranchial body might be the origin of fistula [6, 22]. Lack of an ultimobranchial body in the right neck in mammals is the cause of predominance in the left lobe of thyroid [11, 22].

Typical clinical picture of AST includes neck pain, fever, dysphagia, dysphonia, odynophagia and local erythema and a sensation of increased warmth [2, 12]. Fine-needle aspiration is a useful technique to identify specific pathogen, both aerobic and anaerobic cultures should be obtained [1, 13]. A barium swallow examination is used as an efficient research tool to determine the presence of fistula, with a sensitivity estimated at 80% [4, 5, 8, 14, 15, 17]. Furthermore, other radiology techniques such as CT scanning and thyroid scintigraphy may be useful for diagnostic purposes, especially to assess the extent of infection [1, 18–20]. Neck ultrasonography and sonoelastography may play an undeniable role in acute thyroiditis [21, 23]. Endoscopic examination as an invasive procedure may be helpful to confirm the internal orifice of the fistula, but is not mandatory if barium meal study shows the fistula duct [19].

Treatment of AST due to PSF is based initially on antibiotics, incision, and drainage of the abscess formation. Fistulectomy is acknowledged to be the best radical treatment preventing recurrence [2, 3, 7, 11, 14, 19, 20]. Pathologic detection of the fistula may not always be achieved [19]; a typical photomicrograph shows a cleft lined by stratified squamous epithelium

with a various level of chronic inflammatory infiltration [2, 7, 11, 14, 19, 20].

Conclusions

Frequent recurrence of AST must be considered with the anatomical malformation described as pyriform sinus fistula. In most cases, inter-disciplinary proceedings are necessary for the diagnosis and treatment of AST and PSF. Fistulectomy is established as the treatment of choice and should be done routinely.

References

- Shah SS, Baum SG. Diagnosis and Management of Infectious Thyroiditis. *Curr Infect Dis Rep*, 2000; 2: 147–153.
- Gopan T, Strome M, Hoschar A et al. Recurrent acute suppurative thyroiditis attributable to a pyriform sinus fistula in an adult. *Endocr Pract* 2007; 13: 662–666.
- Dokmetas HS, Koyuncu A, Korkmaz S et al. Two cases of acute suppurative thyroiditis secondary to pyriform sinus fistula. *Saudi Med J* 2008; 29: 142–143.
- Takai SI, Matsuzuka F, Kosaki G et al. Internal fistula as a route of infection in acute suppurative thyroiditis. *Lancet* 1979; 1: 751–752.
- Miyauchi A, Matsuzuka F, Takai S et al. Piriform sinus fistula. A route of infection in acute suppurative thyroiditis. *Arch Surg* 1981; 116: 66–69.
- Himi T, Kataura A. Distribution of C cells in the thyroid gland with pyriform sinus fistula. *Otolaryngol Head Neck Surg* 1995; 112: 268–273.
- 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: 400–405.
- Tucker HM, Skolnick ML. Fourth branchial cleft (pharyngeal pouch) remnant. *Trans Am Acad Ophthalmol Otolaryngol* 1973; 77: ORL368–371.
- Sandborn WD, Shafer AD. A branchial cleft cyst of fourth pouch origin. *J Pediatr Surg* 1972; 7: 82.
- Taylor WE, Myer CM, Hays LL et al. Acute suppurative thyroiditis in children. *Laryngoscope* 1982; 92: 1269–1273.
- Shimazaki T, Yoshida Y, Umento H et al. Two cases of pyriform sinus fistula which required a long time for diagnosis. *Auris Nasus Larynx* 1999; 26: 501–507.
- Rossiter JL. Topf Acute suppurative thyroiditis with bilateral pyriform sinus fistulae. *Otolaryngol Head Neck Surg* 1991; 105: 625–628.
- 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: 415–417.
- 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: 953–956.
- Yamashita H, Noguchi T, Takahashi M. Recurrent cervical abscess due to pyriform sinus fistula. *J Laryngol Otol* 1995; 109: 886–888.
- Yolmo D, Madana J, Kalaiarasi R et al. Retrospective case review of pyriform fistulae of third branchial arch origin commonly presenting as acute suppurative thyroiditis in children. *J Laryngol Otol* 2012; 126: 737–742.
- Inoue K, Kozawa J, Funahashi T et al. Right-sided acute suppurative thyroiditis caused by infectious endocarditis. *Intern Med* 2011; 50: 2893–2897.
- Masuoka H, Miyauchi A, Tomoda C et al. Imaging studies in sixty patients with acute suppurative thyroiditis. *Thyroid* 2011; 21: 1075–1080.
- Seki N, Himi T. Retrospective review of 13 cases of pyriform sinus fistula. *Am J Otolaryngol* 2007; 28: 55–58.
- Hatakeyama H, Homma A, Nagahashi T et al. A study of pyriform sinus fistula cases. *Auris Nasus Larynx* 2001; 28 (Suppl): S139–143.
- Ruchala M, Szczepanek-Parulska E, Zybek A et al. The role of sonoelastography in acute, subacute and chronic thyroiditis: a novel application of the method. *Eur J Endocrinol* 2012; 166: 425–432.
- Zatonski T, Inglot J, Krecicki T. Torbiel boczna szyi. *Pol Merk Lek* 2012; 32: 191, 341.
- Ruchala M, Szczepanek E. Thyroid ultrasund — a piece of cake? *Endokrynol Pol* 2010; 61: 330–344.