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

Łukasz Cieszyński M.D., Department of Endocrinology and Internal Medicine, Medical University of Gdansk, Dębinki St. 7, 80–952 Gdansk, Poland, e-mail: lukaszdoc@wp.pl

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

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 melaninogenic*al. 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 thyreotropin (TSH) with serum free triiodothyronine (FT3).
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 cistern 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 amoxycillin.

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 embryo tract. On the other hand, the spatial maldevelopment 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 pain, fever, dysphagia, dysphonia, odynophagia [2, 7, 8]. PSF is usually associated with infection of the upper respiratory tract. On the other hand, infected patients with PSF required a long time for diagnosis. After 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 the management of acute suppurative thyroiditis. Ultrasound and thyroid scintigraphy may be useful for the diagnosis and treatment of AST and PSF. Fistulectomy is established as the treatment of choice and should be done routinely.

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