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RECURRENT ACUTE SUPPURATIVE THYROIDITIS SECONDARY TO PYRIFORM SINUS FISTULA DUE TO *STAPHYLOCOCCUS SCIURI* WITH SUBCLINICAL HYPERTHYROIDISM IN AN ADULT. A DIAGNOSTIC AND THERAPEUTIC CHALLENGE

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ABSTRACT

Background: Acute suppurative thyroiditis (AST) caused by pyriform sinus fistula (PSF) is a rare condition in adults. PSF is a congenital malformation that can cause an infection of the thyroid gland, usually located on the left side. AST presents with symptoms similar to respiratory infections, which can make it difficult to diagnose. It is essential to consider PFS as a differential diagnosis due to its relationship with the recurrence of thyroid abscesses.

Case report: We report the case of a 32-year-old woman who presented with neck pain and fever, initially mistaken for an upper respiratory infection. After 3 weeks, she was diagnosed with AST with an abscess in the left thyroid lobe. Drainage and initial antibiotic treatment (metronidazole and clindamycin) improved her condition. However, due to non-compliance with medical follow-up, the patient suffered a recurrence of the abscess and required a left thyroid lobectomy. The culture identified multidrug-resistant *Staphylococcus sciuri* and the patient was finally referred for a successful fistulectomy.

Conclusion: AST due to PSF is uncommon in adults, with few documented cases. Initial treatment includes antibiotics and surgical drainage, although recurrence can occur if the risk factor is not eliminated. In this case, failure to comply with medical follow-up contributed to the recurrence. It is crucial to identify PSF as a causative factor early on and to carry out adequate follow-up to avoid serious complications.

KEYWORDS

Suppurative thyroiditis, subclinical hyperthyroidism, pyriform sinus fistula, *Staphylococcus sciuri*

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LEARNING POINTS

- Thyrotoxicosis can occur in up to 42% of cases of acute suppurative thyroiditis, making it less relevant to differentiate it from other causes of thyroiditis.
- In adults with no obvious risk factors, congenital malformations should be considered, especially when the condition affects the left lobe.
- To avoid recurrences, it is necessary to evaluate the pyriform sinus in any age group and patient adherence to medical instructions is essential.

INTRODUCTION

Acute suppurative thyroiditis (AST) caused by pyriform sinus fistula (PSF) in adults is a rare condition, with few cases documented in the medical literature. PSF is a rare congenital malformation that originates in the third or fourth branchial arch, forming an abnormal duct through which bacteria can infect the thyroid gland, causing suppurative thyroiditis. Although the incidence of this condition is low, its clinical presentation can be complex, as the symptoms are often initially confused with upper respiratory infections or benign neck conditions. Therefore, it is essential to consider PSF as a differential diagnosis, especially when thyroid abscesses are located on the left side.

We present a case of AST due to PSF with subclinical hyperthyroidism, complicated by recurrence of the abscess due to *Staphylococcus sciuri*.

CASE DESCRIPTION

The patient was a 32-year-old woman, resident of Mexico, Playa del Carmen, who worked as a chambermaid. She had a history of asthma treated with salmeterol/fluticasone, cholecystectomy 3 years ago. She began to suffer pain on the front of her neck, and due to suspicion of an upper respiratory tract infection, she was given painkillers and antibiotics without improvement. After 3 weeks her left thyroid lobe had enlarged, she had a fever and the pain had returned, so it was decided to hospitalize her. On admission she had a blood pressure of 130/70 mmHg, heart rate of 101 bpm, temperature of 38.5°C and oxygen saturation of 99%. Her body mass index was 25 kg/m^2. Neurological examination results were normal. In the neck, there was a 5 cm nodule on the left side of level VI, painful, erythematous and mobile, as well as enlarged lymph nodes on the same side at level V. Cardiopulmonary findings were normal. The rest of the examination was unremarkable.

Laboratory test results were as follows: venereal disease research laboratory test and human immunodeficiency virus: negative, thyroid stimulating hormone (TSH) 0.178 mIU/l (normal range: 0.49-3.5 mIU/l) and free thyroxine (T4) 1.09 ng/dl (normal range: 0.4-2 ng/dl). A computed tomography (CT) scan of the neck showed a tumour in the left lobe of the thyroid and an ultrasound showed a lesion on the left measuring 2.8×3.5×3.1 cm, suspected of being an abscess. The fine needle aspiration cytology was negative for malignant cells, with lymphocytes and polymorphonuclear cells, compatible with colloid goitre. The conclusion was that the patient had AST. The abscess was drained and antibiotic therapy with metronidazole and clindamycin was administered, with good progress, and she was discharged to outpatient endocrinology consultation after 10 days. In this consultation, a new ultrasound was obtained, reporting no evidence of an abscess but with a 2 mm nodule in the right thyroid lobe with benign characteristics. A fistula of the pyriform sinus was suspected and confirmed with an esophagogram *(Fig. 1)*. The patient was referred to general surgery, however, poor adherence to her follow-up and prolonging the surgical evaluation for a year, resulted in recurrence of the abscess with spontaneous drainage to the oral cavity, requiring readmission to hospital, antibiotics, drainage and left thyroid lobectomy. The culture identified multidrug-resistant *S. sciuri* and the histopathological study reported diffuse nodular hyperplasia, capsular fibrosis with acute chronic inflammation and absence of malignancy. The thyroid function test results were normal (TSH: 1.80

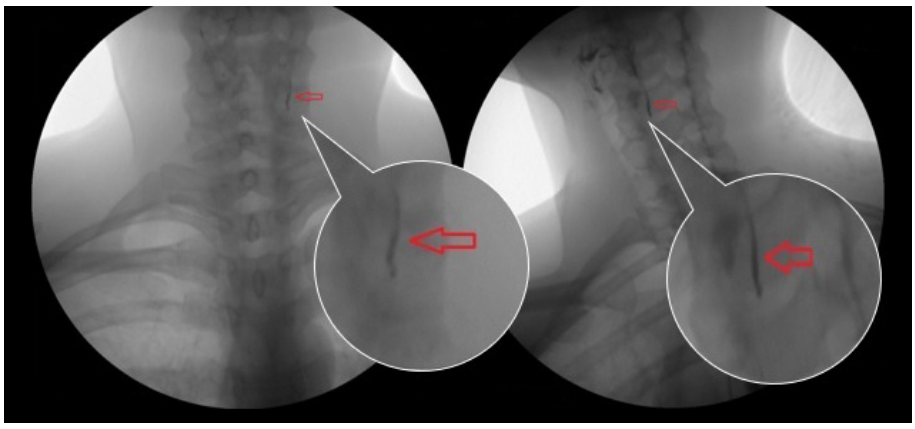


Figure 1. Fluoroscopy esophagogram showing thin leak of contrast material from the inferior apex of the pyriform sinus to the left (red arrow).

mIU/l, free T4: 0.91 ng/dl). The patient's progress in hospital was satisfactory and she was discharged and referred to a specialized centre in Mexico City where the fistulectomy was successfully performed.

DISCUSSION

Acute suppurative thyroiditis (AST) is a rare condition that accounts for between 0.1 and 0.7% of all thyroid diseases[1]. A descriptive review of cases between 2000 and 2020 showed that the average age of patients with AST was 37 years, with a range of 1 to 87 years. There is a predominance of women (54%), which is consistent with our report. However, the cause of AST is unknown. The average time from onset of symptoms to identification of AST is 7 days in bacterial aetiology, 21 days in fungal cases, and up to 30 days in tuberculosis cases[2]. This time may vary depending on the health system, the level of experience, and the degree of clinical suspicion. In our case, the time was 21 days, given that the patient was attended to at a primary level, where the symptoms were confused with an upper respiratory tract infection. For this reason, the time to diagnosis was prolonged until the infectious nodule became evident.

The most frequently documented microorganisms that cause AST are *Staphylococcus aureus* and streptococci (39%), polymicrobial (30%), Gram-negative bacilli (25%), anaerobes (12%) and other microorganisms, such as *Pneumocystis jirovecii*, fungi, *Nocardia* or mycobacteria (15%)[2]. There have even been cases caused by *Echinococcus*[3], to the best of our knowledge this is the first report to identify *S. sciuri*.

The clinical manifestations of AST are: pain (89%), neck swelling, erythema, fever (82%), dysphagia (46%), general malaise and, occasionally, spontaneous drainage of an abscess[2]. The classic literature describes a picture of euthyroidism[3], but currently this occurs in 54% of cases; thyrotoxicosis in 42%, with free T4 levels at twice the normal level in 48%, and hypothyroidism in 5%[2]. The patient presented biochemical thyrotoxicosis (suppressed TSH and normal free T4, without symptoms), which was transient and did not require medical management, which is consistent with the second most frequent manifestation in thyroid function tests in AST.

Given that subacute granulomatous thyroiditis is a similar condition, alterations in thyroid function tests do not allow it to be differentiated from AST. However, the most effective methods are ultrasound alterations and fine needle aspiration of purulent fluid[2].

The initial treatment for AST includes antibiotic therapy and surgical drainage, however, some reports have highlighted percutaneous drainage and washing as an alternative, although its effectiveness has been demonstrated in few cases[4]. Mortality is 5% in bacterial aetiology, 33% in fungal aetiology and 50% in parasitic aetiology[2].

Recurrence occurs when the risk factor persists, so it is necessary to eliminate this factor early to avoid increased healthcare costs, and complications such as abscess recurrence, multi-resistant infection, thyroid lobectomy, sepsis and even death. In our description, the patient's failure to attend medical appointments which delayed the fistulectomy stands out, and this behaviour should also be considered as a risk factor for complications.

The thyroid gland is resistant to infection through multiple mechanisms. However, there are risk factors for infection, such as immunosuppression (27%), anatomical abnormalities (21%), previous thyroid disease (20%), diabetes (13%) and disseminated infections (12%)[2]. It should be noted that, within our diagnostic approach, we ruled out these factors, except for anatomical abnormalities, so the presence of PFS was evaluated and confirmed, this being the second most common cause mentioned in the aforementioned study. This casts doubt on reports about the rarity of this factor, although the authors did not categorize the incidence in adults.

PSF accounts for between 4 and 8% of branchial anomalies. Its association with AST has been documented mainly in children, since 80% of cases manifest in the neonatal stage and childhood. This contrasts with the clinical case we present, given that PSF is found in only 4.1% of adults. These alterations have a predominance on the left side. The fistulas result from the persistence of the pharyngotracheal duct between the superior parathyroid gland and the last branchial arch. There are few reports of PSF in adults, and they have been identified as a cause of recurrence, as happened to our patient, who presented with a recurrent abscess while waiting for fistulectomy. The gold standard for diagnosis is suspension laryngoscopy; however, because it requires general anaesthesia, other diagnostic methods are chosen, such as ultrasonography, CT scan, esophagogram and magnetic resonance imaging. The esophagogram study has a sensitivity of 81.58%, making it the second most effective of the previously mentioned methods[5], which is why we used it as a diagnostic method.

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