

Case Report

Acute Suppurative Thyroiditis Due to Pyriform Sinus Fistula: A Case Report

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Congenital pyriform sinus fistula is an unusual developmental abnormality, but it is the most common cause of acute suppurative thyroiditis or recurrent lateral neck infection in pediatric group. It is often overlooked because of poor diagnostic roles. Further investigations are important for diagnosis and evaluation. The authors herein present a case of pyriform sinus fistula and diagnostic roles to confirm the diagnosis.

Keywords: Acute suppurative thyroiditis, Pyriform sinus fistula

J Med Assoc Thai 2010; 93 (3): 388-92

Full text. e-Journal: <http://www.mat.or.th/journal>

Acute suppurative thyroiditis is an uncommon infectious disease, especially in the pediatric group. It occurs due to bacterial causes or fungus. It rarely occurs in healthy children. Congenital or acquired anatomical abnormalities, previous diseases of the thyroid gland, immunocompromised, or debilitated patients usually predispose to infectious events⁽¹⁾.

Congenital pyriform sinus fistula is a branchial pouch abnormality. The remnant tract originates from the third or fourth branchial cleft anomaly^(2,3). The tract pathway begins from the apex of the pyriform sinus, passes through or beside the thyroid gland anterocaudally, and terminates in perithyroid tissue^(3,4). It is an extreme diagnostic and therapeutic challenge.

Case Report

A 4-year-old previously healthy girl presented with 2-week history of pharyngitis and flu-like symptoms. She was treated as viral upper respiratory tract infection. Her symptom partially improved with persistent low-grade fever. She complained of left-sided neck pain and swelling 10 days later. She got routine vaccination and had no previous history of neck injury or cervical irradiation. The family history was negative for thyroid disease or cancer. On

admission, her temperature was 36.9°C. Physical examination revealed a large firm, erythema, warm, tender neck mass to the left of the midline in the region of the thyroid gland and relatively fixed. It did not move with swallowing. The trachea was mildly shifted to the right. There was no cervical lymphadenopathy. She was clinically euthyroid. Laboratory investigations revealed a hemoglobin value of 9.6 g/dl and a leukocyte count of 15,800 with 66% polymorphonuclear cells. The thyroid function tests were within the normal range. The sonographic examination performed with a high-frequency (12-MHz) probe disclosed the ill-defined non-homogeneous hypoechoic lesion involving the left lobe of the thyroid gland and perithyroidal area, but could not depict a sinus or fistulous tract. Increased vascular perfusion at this area was noted (Fig. 1). The parenchymal structure of the right lobe was intact. There were several small left lateral cervical lymph nodes. Unilateral thyroiditis of the left lobe with extension of inflammation to soft tissue of left neck was diagnosed. Because unilateral thyroiditis, especially left side, is unusual in healthy children, pyriform sinus fistula, as the underlying cause must be excluded and computed tomography (CT) of the neck was requested.

The CT revealed a multi-lobulated hypo-attenuation mass in the left-sided of the neck with associated surrounding mild enlargement of the left sternocleidomastoid muscle, swelling of the left strap

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muscles, and adjacent skin thickening. Slight loss of high attenuation of the affected left lobe of thyroid gland after contrast enhancement was also evident. Additionally, the left pyriform fossa was deformed by the adjacent soft tissue inflammation (Fig. 2). There were multiple small lymph nodes along bilateral jugular chain. Left pyriform sinus fistula was still highly suspected even though the sinus tract between left pyriform sinus and left lobe of thyroid could not be demonstrated by CT.

Incisional and drainage of the left-sided neck mass was performed. The intraoperative showed necrotic tissue at the left-sided neck mass, measuring approximately 3-cm with minimal pus. The microscopy revealed acute inflammatory exudates with granulation

tissue and positive culture for *Streptococcus intermedius*.

The patient was treated with intravenous cloxacillin at a dose of 100 mg/kg/day for about three days and then increased dose of cloxacillin at a dose of 150 mg/kg/day for cover anaerobe bacteria.

Barium swallowing was obtained 18 days after resolution of her illness episode for detection of congenital pyriform sinus fistula, which was thought to be the cause of this infection. The patient will have chronic recurrent left thyroiditis if this anomaly was not recognized. The present study demonstrated a thin sinus tract, approximately 1.1 cm in length, originated from the inferior aspect of the left pyriform sinus extending inferiorly into the soft tissue on the left-sided

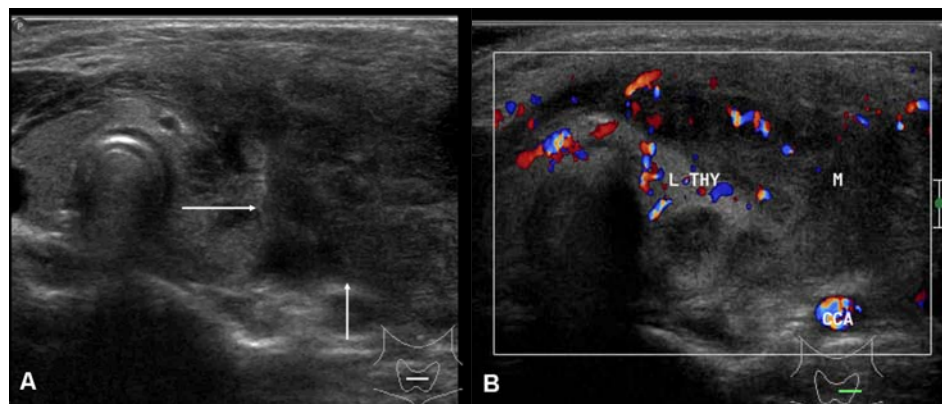


Fig. 1 Transverse ultrasound shows an ill-defined non-homogeneous hypoechoic lesion (arrows) at the left lobe of thyroid and perithyroidal area (A) with increased vascular perfusion (B). L THY = left lobe of thyroid gland, M = mass lesion and CCA = left common carotid artery

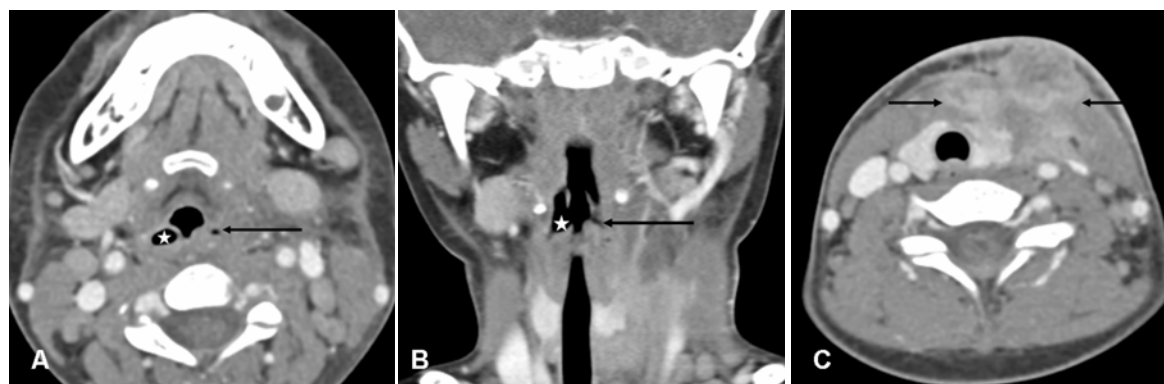


Fig. 2 A, B: Axial (A) and coronal (B) CT shows deformity of the left pyriform fossa (arrow) by the adjacent soft tissue swelling, compared to the right pyriform side (white star). C: Axial CT (lower level from A) shows multilobulated hypoattenuation mass in left perithyroid region (arrows) with slightly decreased high density of the left thyroid lobe

of the neck, consistent with a persisting pyriform fossa sinus. No significant tracheal narrowing was depicted (Fig. 3).

Twenty days after barium swallowing, she underwent elective surgery for left hemithyroidectomy and fistulectomy. The patient had no recurrence of thyroiditis after surgery.

Discussion

Congenital pyriform sinus fistula must be considered as a possible diagnosis in patients presenting with acute and unilateral suppurative thyroiditis, neck mass, or recurrent lateral neck infection/abscess^(5,6). The clinical characterizations of acute suppurative thyroiditis are abrupt onset of painful swelling in the left-sided of thyroid region with fever, occasionally following upper respiratory tract infection⁽⁷⁾. It is usually unilateral (mostly left-sided). It is often undiagnosed until complications such as acute suppurative thyroiditis, deep neck infection, or mediastinal abscess result. The age at onset is between 2 and 12 years^(7,8). Chronic recurrent thyroiditis will occur if not correctly diagnosed and surgically removed.

Direct visualization of a sinus or fistulous tract in pyriform sinus is the diagnostic⁽⁶⁾. Additionally, the proper technical issues or guidelines about diagnostic work-up are important role to demonstrate.

Plain roentgenograms of the neck are non-specific. Miyauchi et al showed soft tissue swelling on

the left-sided neck, causing trachea shifting to the right, and a few cases had gas shadow and a gas-filled level in the swelling area⁽⁹⁾.

The imaging in our patient followed the pattern. Ultrasound (US) is often the initial imaging modality used for evaluating pediatric patients who presented with neck mass. The characteristic findings are an ill-defined hypoechoic area or heterogeneous mass involving the upper two thirds of the left lobe of the thyroid gland or left-sided perithyroid area with or without effacement of the intervening plane of the left thyroid lobe^(2-4,7), which are similar findings in our patient. However, they were non-specific for the causes of acute suppurative thyroiditis. Sometime, they can demonstrate internal debris content or gas bubbles. US are limited to depict a sinus or fistulous tract and hypopharyngeal lesion.

The barium swallowing is easy and can demonstrate the opening of the sinus or fistulous tract originating from the left pyriform sinus^(3,4,10). In our case, barium swallowing was done 18 days after the inflammation was subsided and well demonstrated the pyriform sinus fistula. In the resting stage of disease, the barium swallowing has 80% sensitivity for assessing the presence of an anomalous tract. However, false-negative results may occur due to the edematous soft tissue causing obliteration or closure of the tract⁽⁸⁾. Skuza et al recommended barium swallowing to be done about two months after an acute infectious episode⁽¹¹⁾.

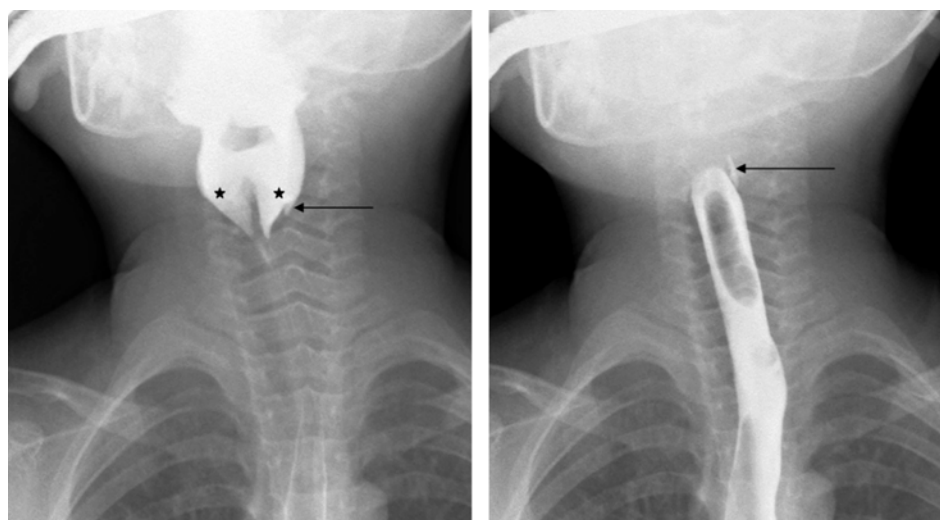


Fig. 3 Barium swallowing demonstrates thin sinus tract (arrow), originates from inferior lateral wall of the left pyriform sinus, extending inferiorly into the soft tissue on the left-sided of neck. Pyriform sinuses = black stars

CT or magnetic resonance (MR) imaging is often the technique of choice to assess deep neck infection, evaluation of the extent of infection, and complications. These show abnormal soft tissue swelling with enhancement along the course of the tract, abscess formation, pyriform sinus deformity due to adjacent soft tissue edema, swelling, and loss of the normal attenuation of left thyroid lobe⁽²⁾. However, CT is superior in detecting the air density at the extrapharyngeal area, the sinus, or fistulous tract, or in the affected thyroid gland. It is an available and fast technique⁽⁴⁾. Modified Valsalva maneuver during the CT may be effective to demonstrate the pharyngoathyroid communication⁽¹²⁾. In our case, the pyriform sinus fistula could not be demonstrated by CT.

Surgical exploration with complete excision of the entire sinus or fistula tract is the treatment of choice^(8,13).

The authors report this case even though many cases diagnosed by barium swallowing function had been reported in the world literatures because of its unique clinical symptoms. Signs should alert clinicians to think of this anomaly and perform a correct diagnostic.

Conclusion

Acute suppurative thyroiditis associated with pyriform sinus fistula is an unusual disorder, but has the characteristic clinical manifestation. The clinical awareness is very important. The diagnosis should be suggested whenever an inflammatory process or abscess is present in the unilateral lobe of thyroid, lower neck along course of the pyriform sinus fistula, especially on the left-sided.

Further investigations are important for diagnosis and evaluating the extent of the lesion as well as its complications. Ultrasound and CT demonstrate neck infection but may be limited to depict the sinus or fistula tract. The most important sign is obtained by barium swallowing, which can delineate the fistula and its extension prior to surgical treatment.

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การติดเชื้อของต่อมไทรอยด์จากสาเหตุ *pyriform sinus fistula*: รายงานผู้ป่วย 1 ราย

มยุรีวรรณ ตะเพย, รัตนพร พรกุล

Pyriform sinus fistula เป็นความผิดปกติตั้งแต่กำเนิดที่พบบได้น้อย ดังนั้นแพทย์ผู้ตรวจจึงมักไม่ค่อยนึกถึง แต่ภาวะนี้มักจะเป็นสาเหตุของการติดเชื้อที่ต่อมไทรอยด์ โดยเฉพาะการติดเชื้อที่เกิดขึ้นซ้ำ ๆ ในกลุ่มผู้ป่วยเด็ก ผู้เขียนได้รายงานผู้ป่วย 1 ราย ที่มาตรวจด้วยอาการปวด บวม และมีก้อนที่คอ รวมถึงถึงความสำคัญ และหลักในการส่งตรวจพิเศษทางรังสีที่ช่วยในการวินิจฉัยผู้ป่วย
