

Diagnosis of Acute Suppurative Thyroiditis Secondary to Pyriform Sinus Fistula by Local Injection of Ultrasound Contrast Agent

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Abstract: Pyriform sinus fistula (PSF) is a rare congenital branchial anomaly that commonly found in children. It can result in recurrent cervical infection or acute suppurative thyroiditis (AST). Confirming the fistula tract is the key point for the diagnosis of PSF. Ultrasound plays an important role in the diagnosis of PSF and its complications. We examine a case of AST secondary to PSF by ultrasonography, and successfully demonstrate its fistula through local injection of ultrasound contrast agent into the thyroid abscess cavity. This is the first report that confirms a fistula tract through local injection of ultrasound contrast agent, and it can serve as an accurate modality for the fistula detection of PSF.

Keywords: Pyriform sinus fistula; Acute suppurative thyroiditis; Local injection; Contrast agent; Ultrasound

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Pyriform sinus fistula (PSF) is a congenital branchial anomaly, resulting from failure of obliteration of the third or fourth branchial pouch. It emerges from the pyriform sinus and usually involves the lateral lobe of the thyroid gland [1]. This anomaly usually presents with recurrent cervical infection or acute suppurative thyroiditis (AST) in children, mostly on the left side of neck [2]. The definitive diagnosis of PSF requires the detection of a fistula. Ultrasound (US) has been considered to have a minor role in diagnosing PSF, because of its limited ability to detect a laryngohypopharyngeal lesion [1]. In this case study, we demonstrate the ability to visualize a fistula tract by injecting ultrasound contrast agent into the thyroid abscess cavity, and provide a new method for the detection of PSF.

Case Report

A 14-year-old boy presented with left neck pain, swelling, and intermittent fever for 2 weeks. He was initially diagnosed with subacute thyroiditis and was

prescribed antibiotics and steroids at another hospital. However, the treatment was not effective, and his symptoms aggravated 2 days later. For accurate diagnosis and treatment, he was admitted to our hospital. The boy had a previous history of subacute thyroiditis at the age of 7 years and received steroid therapy for more than 1 month. After physical examination, we observed that the patient had a fever of 38.7°C, and his left neck was swollen with local dermal erythema. The laboratory findings were as follows: a white blood cell count of $19.58 \times 10^9/L$, a Neutrophil count of 78.6%, a C-reactive protein level of 84 mg/L, an erythrocyte sedimentation rate of 65 mm, and normal thyroid function and thyroid antibody titer levels (including TPO-Ab and TG-Ab). Cervical ultrasonography showed an ill-defined, heterogeneous, and hypoechoic mass measuring $5.0 \times 3.4 \times 3.1$ cm in the left lobe of the thyroid gland (Fig. 1A). Color Doppler Flow Imaging (CDFI) revealed peripheral blood flow in the mass. Moreover, multiple enlarged lymph nodes were observed in the left neck. Acute suppurative thyroiditis (AST) with abscess formation was suspected. To validate our suspicion, ultrasound-guided aspiration

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of the abscess was performed (Fig. 1B, 1C). Culture was positive for Gram-positive cocci, Gram-negative bacilli, and anaerobic bacteria. Diagnosis of AST was concluded.

Bacterial infection rarely occurs in the thyroid gland due to its dense capsule, abundant blood supply, and lymphatic drainage. Recurrent AST in a child strongly indicates the existence of an underlying pyriform sinus fistula (PSF). Confirming the fistula tract and its opening is the key point for the diagnosis of PSF. Unfortunately, barium esophagography did not reveal the presence of a fistula in this case. So, after the aspiration procedure, we injected ultrasound contrast agent into the abscess cavity

of the left thyroid gland (Fig. 2A, 2B). Also, we asked the patient to swallow several times after injection, then a dynamic flow of contrast agent was observed into the posterolateral aspect of the thyroid cartilage inferior edge (Fig. 2C, 2D), which indicated a sinus tract from the pyriform sinus. After his local infection was controlled, the patient received surgery to remove the fistula extending from the pyriform sinus to the lower border of the thyroid gland. Postoperative pathology findings supported the diagnosis of PSF.

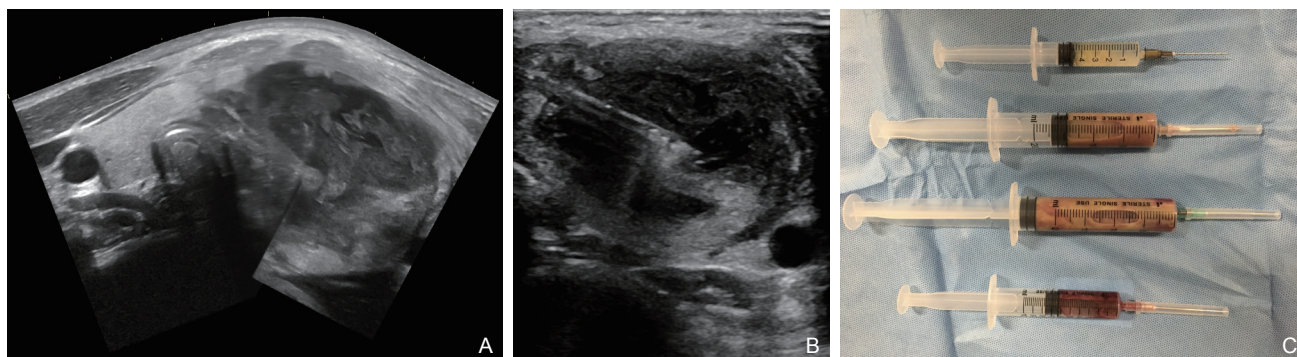


Figure 1 (A) Panoramic image showing a hypoechoic mass in the left lobe of the thyroid gland; (B) US-guided aspiration of the lesion was carried out; (C) Approximately 50 mL of pus was aspirated.

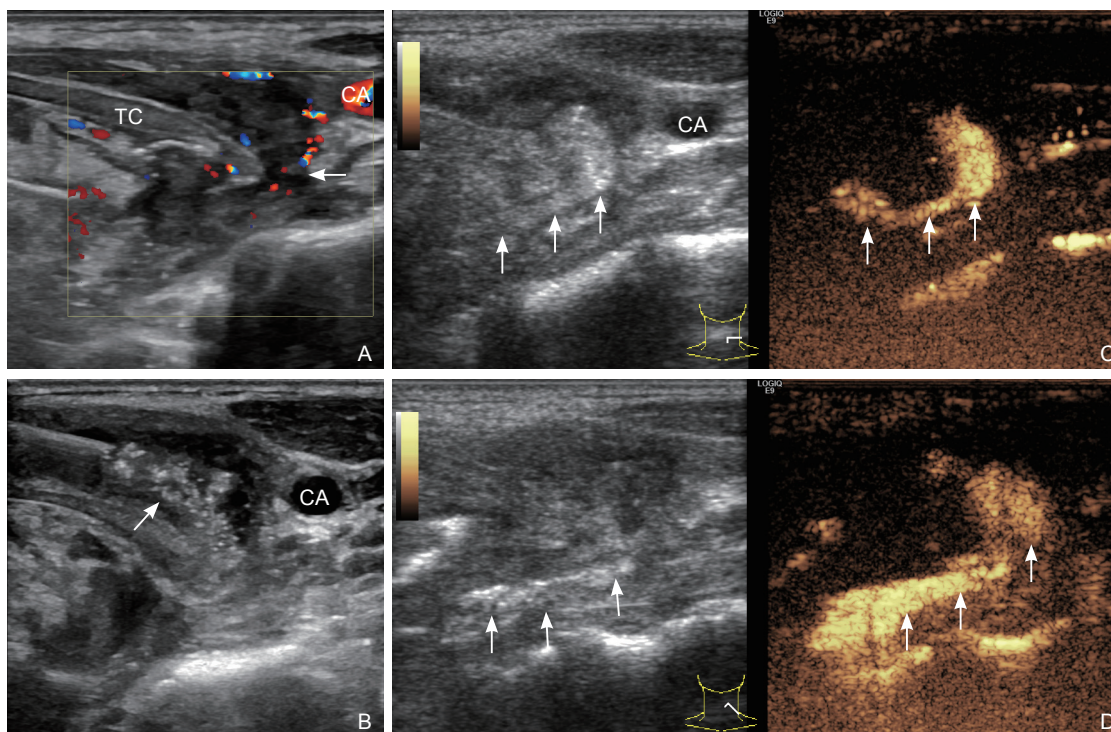


Figure 2 (A) The transverse image showing an abscess emerging from the posterolateral aspect of the thyroid cartilage (TC), which indicated that the abscess started from the pyriform sinus; (B) Injection of ultrasound contrast agent into the abscess. The images showing the fistula (C and D). TC, Thyroid cartilage; CA, Common carotid artery.

Discussion

Generally, it is believed that PSF results from

failure of obliteration of the third or fourth branchial pouch. Persistence of the thymopharyngeal duct of the

third pouch, frequently passing through or adjacent to the thyroid gland, is thought to be the most suitable embryologic explanation of PSF [3]. At the 6th week of gestation, the primordia of thymus arises from the ventral part of the third branchial pouch and elongates caudally. As the thymic primordia migrates caudally, there is a thymopharyngeal duct connecting the pharynx and thymic primordia. The duct is usually obliterated after the fusion of the thymic tissues of either side. Failure of obliteration of the thymopharyngeal duct may leave a fistula extending to the pharynx [3, 4]. Clinical presentation of PSF depends on whether there is a cervical infection secondary to the fistula. If no infection is present, there will be no symptoms. However, the fistula acts as a pathway for the propagation of bacteria in the upper respiratory tract, which can lead to recurrent cervical infections, mainly on the left side. It commonly presents as an inflammatory mass in the neck, often involving the thyroid gland. Early diagnosis and surgical removal of the fistula tract is the key point in preventing recurrent cervical infections.

The diagnosis of PSF is challenging. The identification of the fistula tract and its opening is important in confirming an underlying PSF involving the thyroid gland. Barium esophagography and laryngoscopy are commonly performed to validate the presence of a fistula opening [1]. However, the former must be performed approximately 4–6 weeks after the acute inflammatory stage. If performed too early, it will present false-negative results because the oedematous wall of the tract may not allow barium to pass [2]. Furthermore, barium esophagography is poorly tolerated in pediatric patients. Laryngoscopy is an invasive procedure and requires general anesthesia [1]. Besides, barium esophagography and laryngoscopy cannot evaluate the localization and distribution of inflammation. Although computed tomography scanning has high sensitivity in identifying PSF, radiation exposure limits its clinical application [1]. US is the primary modality in evaluating neck masses, and therefore it plays an inevitable role in the diagnosis of PSF. During the early acute inflammatory stage, US can easily identify the inflammatory process associated with the thyroid gland, and it can be performed repeatedly during the management of infection. If necessary, ultrasound-guided aspiration of the abscess can be performed. It will relieve compression on the airway caused by the abscess and will contribute to targeted antibiotic treatment. In terms of the demonstration of PSF and its opening, some reports found that US is less valuable in demonstration of the fistulous tract. Dating back to 2003, H.-K. Wang had successfully demonstrated the tract of PSF by using the trumpet maneuver (or so-called modified Valsalva

maneuver), which instructs the patient to exhale against pursed lips in order to distend the pyriform sinus. However, in clinical practice, the trumpet maneuver is difficult to carry out for an infant or a very young child [5]. Even though kids are capable of doing trumpet maneuver properly, compression from the cervical abscess may limit air bubbles into the fistulous tract, which might still fail to demonstrate the PSF. In this case, we finally identified the existence of a fistula and its anatomical path by injecting ultrasound contrast agent into the abscess cavity. Therefore, contrast-enhanced US has unique advantages for the evaluation of inflammatory process and the diagnosis of PSF. To our knowledge, this is the first report that confirms a PSF via local injection of ultrasound contrast agent. As an innovative method, the injection of ultrasound contrast agent into the cervical lesion is a sensitive method in identifying a fistula.

In the early inflammatory stage with no abscess formation, an ill-defined hypoechoic area involving the thyroid gland is the most common finding on US, which may lead to a misdiagnosis of subacute thyroiditis. AST secondary to PSF must be distinguished from subacute thyroiditis, as steroids commonly used for the treatment of subacute thyroiditis will adversely affect AST [6]. AST has left-side predominance, whereas subacute thyroiditis often successively involves both sides. Several reports, which are about PSF presenting with AST and recurrent neck infection, have been published. However, the PSF across the thyroid gland can manifest as an incidental thyroid nodule mimicking thyroid cancer when there is no infection [7]. Air bubbles from the pyriform sinus mimic the internal hyperechogenic foci on US, which can be confused with microcalcification of papillary thyroid carcinoma, particularly in cases with multiple hyperechoic foci. If fine-needle aspiration is performed under this circumstance, infection will easily be developed. Several US findings may be indicative of a PSF in patients presenting with suppurative thyroiditis or a thyroid nodule. First, PSF has a left-side predominance. Second, it presents as a hypoechoic tubular lesion emerging from the posterolateral aspect of the thyroid cartilage and crossing the thyroid gland. Third, movement of hyperechoic foci in response to US probe compression or during swallowing can help differentiate a PSF from a thyroid nodule [1].

Conclusion

Congenital PSF is the most common cause of AST in children. Recurrent development of suppurative thyroiditis or neck infection, particularly in the left side, is the most important sign of an underlying PSF. US plays an increasingly important role in the diagnosis

and management of AST secondary to PSF, due to its widespread availability and safety. Furthermore, injection of US contrast agent into the abscess is an effective modality to detect PSF.

Supplement

Video. Dynamic flow of contrast agent was observed into the posterolateral aspect of the thyroid cartilage inferior edge.

Conflicts of Interest

The authors have no conflict of interest to declare.

References

- [1] Ahn D, Lee GJ, Sohn JH. Ultrasonographic characteristics of pyriform sinus fistulas involving the thyroid gland. *J Ultrasound Med* 2018; 37: 2631-2636.
- [2] Yolmo D, Madana J, Kalaiarasi R, Gopalakrishnan S, Kiruba Shankar M, Krishnapriya S. Retrospective case review of pyriform sinus fistulae of third branchial arch origin commonly presenting as acute suppurative thyroiditis in children. *J Laryngol Otol* 2012; 126: 737-742.
- [3] Thomas B, Shroff M, Forte V, Blaser S, James A. Revisiting imaging features and the embryologic basis of third and fourth branchial anomalies. *AJNR Am J Neuroradiol* 2010; 31: 755-60.
- [4] Benson MT, Dalen K, Mancuso AA, Kerr HH, Cacciarelli AA, Mafee MF. Congenital anomalies of the branchial apparatus: embryology and pathologic anatomy. *Radiographics* 1992; 12: 943-960.
- [5] Wang HK, Tiu CM, Chou YH, Chang CY. Imaging studies of pyriform sinus fistula. *Pediatr Radiol* 2003; 33: 328-333.
- [6] Masuoka H, Miyauchi A, Tomoda C, Inoue H, Takamura Y, Ito Y, et al. Imaging studies in sixty patients with acute suppurative thyroiditis. *Thyroid*. 2011; 21: 1075-1080.
- [7] Kim SC, Kim JH, Won JK, Chung EJ. Asymptomatic intrathyroidal pyriform sinus fistula mimicking thyroid cancer: A case report and literature review. *Medicine (Baltimore)* 2018; 97: e0488.