

# Asymptomatic intrathyroidal pyriform sinus fistula mimicking thyroid cancer

## A case report and literature review

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### Abstract

**Rationale:** There have been many reports of non-thyroidal lesions which can be mistaken for thyroidal lesions on ultrasound (US) examination. However, it is not known that pyriform sinus fistula (PSF) can manifest as an incidental thyroid nodule and cause serious complication on fine-needle aspiration (FNA).

**Patient concerns:** We present a 34-year-old man with PSF incidentally detected on US. US examination showed hypoechoic nodule with several bright echogenic spots at the uppermost part of left thyroid gland. With the suspicion of thyroid cancer, although there would have been some morphologic changes between the 2 US examinations, FNA was performed.

**Diagnoses:** Cytologic specimen revealed some clusters of ciliated columnar cells mixed with inflammatory and lymphoid cells. On computed tomography (CT) before FNA, there were tiny air bubbles within the thyroid nodule. Laryngoscopy revealed fistula originating from the pyriform sinus.

**Interventions:** After FNA, he had to undergo tracheostomy and removal of abscess due to infectious complication.

**Outcomes:** The deep neck abscesses and infections were controlled after the treatment. At 1 year after FNA, successful chemocauterization with 40% trichloroacetic acid solution was performed for PSF found on laryngoscopy.

**Lessons:** PSF can manifest as an incidental thyroid nodule mimicking thyroid cancer. Special care should be taken when FNA is planned for the nodule with air foci and morphologic changeability at the uppermost part of left thyroid gland.

**Abbreviations:** FNA = fine-needle aspiration, PSF = pyriform sinus fistula, US = ultrasound.

**Keywords:** fine needle aspiration, pyriform sinus, pyriform sinus fistula, thyroid cancer, thyroid nodule

## 1. Introduction

Ultrasonography (US) plays a key role in the diagnosis and management of thyroid nodules and US guided fine-needle aspiration (FNA) is primarily recommended for the further evaluation of thyroid nodules.<sup>[1]</sup> Along with increase of use of high resolution US, there have been many reports of various non-thyroidal lesions including Killian–Jamieson diverticulum and paraesophageal air cyst, which can be mistaken for thyroidal lesions on US.<sup>[2–4]</sup> In most literature, pyriform sinus fistula (PSF) has been presented with the complication cases of acute

suppurative thyroiditis and recurrent deep neck infection and emphasized as an important infection source, especially in children.<sup>[5,6]</sup> Herein, we report the case of asymptomatic intrathyroidal PSF mimicking thyroid cancer on US, which entrapped into FNA that could cause serious infectious complication.

## 2. Case report

This study was approved by the institutional review board of Seoul National University Hospital, and the requirement for informed consent was waived due to its retrospective nature.

A 34-year-old man was referred for an incidental left thyroid nodule. The results of thyroid function tests and complete blood count were within normal ranges. The US demonstrated a 1.1 cm nodule at the uppermost part of left thyroid gland. The nodule showed mild hypoechoogenicity, irregular shape, nonparallel orientation, ill-defined margin, and many bright echogenic spots without acoustic shadowing on US (Fig. 1A).<sup>[7]</sup> Between the 2 US exams with 7 months interval, there were some unrecognized changes in the shape and number of internal bright echogenic foci of the nodule (Fig. 1B). In addition, there was 1.2 cm low-attenuating nodule with air bubble at the uppermost part of left thyroid gland on chest CT (Fig. 1C). Nevertheless, the FNA operator could not notice these findings and FNA was performed under the impression of thyroid cancer. The cytologic specimen revealed many ciliated columnar cells in the background of inflammatory and lymphoid cells. Unfortunately symptoms of neck pain, swelling, fever, and dyspnea developed sequentially

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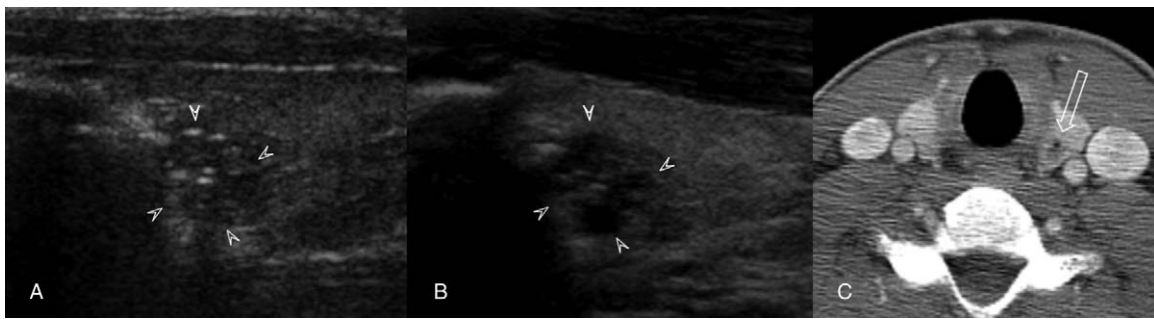
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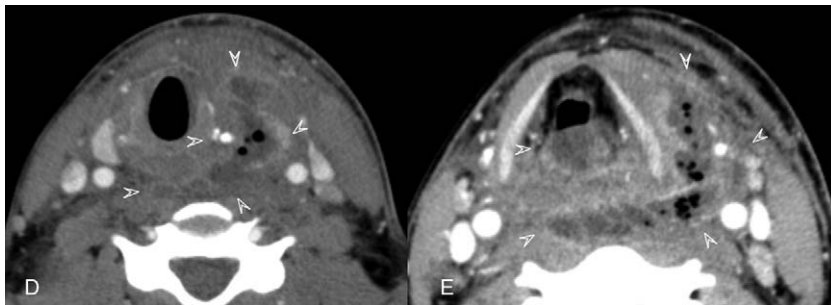
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**Figure 1.** A longitudinal US image (A) shows a nodule (arrowheads) with mild hypoechogenicity, irregular shape, nonparallel orientation, ill-defined margin, and bright echogenic spots without acoustic shadowing in the uppermost part of left thyroid gland. On longitudinal US images (B) obtained 7 months later, there are some changes in the shape of the nodule (arrowheads) and in the number of its internal bright echogenic foci. On chest CT scan (C), there is a 0.2 cm air bubble (white arrow) within a 1.2 cm low-attenuating nodule at the uppermost part of left thyroid gland. CT=computed tomography, US=ultrasound.



**Figure 2.** Three days after the US-guided fine needle aspiration, CT images depict an abscess (arrowheads) involving left upper thyroid gland, adjacent perithyroid tissue, and retropharyngeal space along with diffuse swelling of the hypopharynx and supraglottic larynx. CT=computed tomography, US=ultrasound.

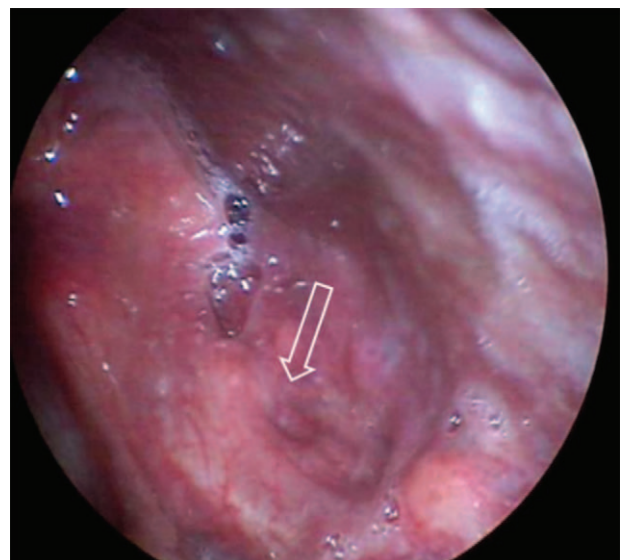
during 3 days after FNA and aggravated in spite of oral antibiotics. When he visited emergency room at 3 days after FNA, the white blood cell count was elevated ( $17,400/\mu\text{L}$ ) and computed tomographic (CT) scan revealed abscess involving left upper thyroid gland, adjacent perithyroid tissue and retropharyngeal space along with diffuse swelling of hypopharynx and supraglottic larynx (Fig. 2). Because of the rapid progression of respiratory distress, he had to undergo tracheostomy and the subsequent incision and drainage of abscess. At 1 year after FNA, successful chemocauterization with 40% trichloroacetic acid solution was performed for PSF found on laryngoscopy (Fig. 3).

### 3. Discussion

Although bacterial infection rarely occurs in thyroid gland due to its rich blood supply and lymphatics and thick fibrous capsule, when PSF exists, the fistula tract can act as pathway for the infection propagation to the thyroid gland.<sup>[5]</sup> As most PSF cases manifest as an infection, it is difficult to interfere with asymptomatic PSF. We firstly report that asymptomatic PSF can manifest as a thyroid nodule and undergo FNA on the clinical scenario.<sup>[5,6]</sup> Although the embryologic origin is controversial, PSF appears to be a remnant originates from the failure of the third or fourth branchial pouches to atrophy and perish in utero, which results in the sinus tract that lie in close proximity to, or inside, the thyroid gland.<sup>[8]</sup>

Until now, there were some reports of intrathyroidal branchial cleft or cleft-like cyst that manifested mainly as cystic lesions.<sup>[9]</sup> However, they were different from our case in the fact that they

were lined by squamous epithelium, and they were not usually located in uppermost part of left thyroid gland. In addition, the connection to the pyriform sinus was not reported in previous studies. Under the name of intrathyroidal branchial cleft sinus, Kim et al<sup>[10]</sup> reported a similar case of this study, which presented



**Figure 3.** A laryngoscopy obtained 1 year later depicts a fistulous opening (white arrow) at the left pyriform sinus.

the thyroid nodule with the suspicious malignant features on US but FNA could be avoided because the lesion changed morphologically during FNA procedure and subsequent CT showed air tract between pyriform sinus and the thyroid lesion.

Followings can be the clues for intrathyroidal PSF. First, the lesions are located in the uppermost part of the thyroid gland. The predominance of left-sided location up to 93% in PSF may be related to the embryologic asymmetry in the persistence of a patent thymopharyngeal duct.<sup>[5,6]</sup> Uppermost location within the thyroid gland can be explained by anatomic proximity to pyriform sinus. Second, air bubbles from pyriform sinus seem to make the internal bright echogenic foci on US. Because internal echogenic spots from air bubbles can be confused with microcalcifications, they may be the important US features in which intrathyroidal PSF can be misregarded as thyroid cancer. Air foci depicted on US or CT seem to be an important clue for the diagnosis of various intrathyroidal structure which communicates with laryngo-pharyngo-esophageal structures.<sup>[2-4]</sup> In the previous series of PSF complicated by neck infections, foci containing air were noted as bubbles or tracts on 9 out of 17 cases.<sup>[5]</sup> Third, the morphologic changeability can be noted between US scans or even during US scan. It can be explained by movability of the extended pharyngoesophageal structure and internal air bubbles. Among the various non-thyroidal lesions mimicking thyroid nodules, Killian–Jamieson diverticulum showed similar features in the aspects of left sided predominant location, foci containing air, and morphologic changeability.<sup>[2]</sup> However, uppermost location of left lobe without connection to esophagus can be an essential clue for differentiating intrathyroidal PSF from Killian–Jamieson diverticulum.

#### 4. Conclusion

PSF can manifests as an incidental thyroid nodule with high suspicious malignant features on US. Because FNA for this lesion can cause a disaster, special care should be taken in the case of the thyroid nodule with air foci and morphologic changeability, in the uppermost part of left thyroid gland.

#### Author contributions

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**Writing – original draft:** Soo chin Kim, Ji-hoon Kim.

**Writing – review & editing:** Soo chin Kim, Ji-hoon Kim.

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