CASE REPORT

Right-sided pyriform sinus fistula in an adult

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Pyriform sinus fistula is important despite its rarity, as it can induce a recurrent neck abscess. Most of the reported cases occur in children and the majority affect only the left side. We report a patient with a pyriform sinus fistula of the right neck in an adult, which was successfully treated by surgery. The aetiology of this entity is also discussed herein.

Introduction

Pyriform sinus fistula (PSF) is notorious for its proclivity to cause recurrent neck abscess or suppurative thyroiditis.[1] Mostly they occur on the left side. Hatabu et al[2] stated that a recurrent right neck abscess virtually excludes the possibility of PSF due to its predominance on left neck.

We report a case of PSF on the right neck in an adult patient, which was complicated by neck abscess formation. The PSF was promptly diagnosed and surgical treatment implemented without delay. This report serves to raise clinical awareness of this entity and its possible aetiology.

Case report

In August 2010, a 29-year-old woman with good past health presented to us because of right neck pain for a week. She had a sore throat and fever, and had developed hoarseness and dysphagia. Her problem did not improve following antibiotic treatment prescribed by a general practitioner. On physical examination, an extremely tender diffuse swelling was found in the right anterior neck. Computed tomography (CT) disclosed a 4.0 x 2.7 x 2.3 cm abscess between the hypopharynx and upper part of right thyroid lobe (Fig 1). However, the epicentre of the abscess was outside the thyroid gland. Flexible endoscopy showed that both vocal cords were mobile but the right pyriform sinus was swollen. The patient was treated with incision and drainage of the abscess. The pus grew *Streptococcus milleri* and *Bacteroides* species. The acute infection subsided after drainage and antibiotic treatment.

Based on the location of the abscess cavity, endoscopic findings and absence of an evident cause, the possibility of right PSF was suspected even though it is extremely rare. A barium swallow done 3 weeks later revealed a sinus tract of 1.8 cm which arose from the apex of right pyriform sinus (Fig 2a). The ultimate diagnosis was right PSF.

To prevent further neck abscess formation, surgical removal of the right PSF was carried out in December 2010. Under general anaesthesia, upper fiberoptic endoscopy revealed the internal orifice of the sinus tract at the posterior part of right pyriform fossa near the apex (Fig 2b). Methylene blue solution was introduced through the endoscope cannula via the working channel of the fiberoptic endoscope, and was injected into the sinus tract orifice to facilitate the subsequent identification of tract during the operation. The transverse right neck scar at level of the cricoid cartilage made during previous abscess drainage was incised. Scar tissue was present at the right upper thyroid pole and was dissected until the blue-stained fistula tract was identified. The tract led upward to the right pyriform apex and traversed the inferior pharyngeal sphincter muscle at the posterior border of thyroid cartilage. Complete extirpation of the tract was carried out. The small defect at the right pyriform fossa was closed with absorbable sutures. The right thyroid lobe was also removed as it was densely adherent to the sinus tract and the strap muscle. The patient recovered from the operation uneventfully. There has been no further attack of neck abscess after the surgery. Pathological examination showed that the tract was lined by stratified squamous epithelium, and immunochemical staining for calcitonin was negative.

Discussion

Because of low levels of awareness, many patients with PSF are diagnosed late despite

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右邊梨狀靜脈竇廔管成人病例

梨狀靜脈竇廔管雖罕見但不可小窺，因它可導致週期性脖膿瘍。大部份病例報告皆涉及兒童患者和影響左脖為主。本文報告一名成人患者的右脖出現梨狀靜脈竇廔管並以手術成功治療，並探討此病的病源學。

repeated attacks of neck infection.[3] Moreover, it is a rare condition occurring on the left side and affecting children. Miyauchi et al[1] reported a series of 15 cases, all on the left side. Likewise, Seki and Himi[4] described 13 cases of PFS and all of which were on left side. However, PSF on right side do exist. In the literature reviewed by Godin et al,[5] two (7%) of right PSF lesions in the 28 patients were reported. Our patient adds one more case of right PSF. For any patients with right perithyroid abscess, right-sided lesions should be considered akin to their left-sided counterparts. Contrast swallow study after the resolution of the acute infection should be undertaken to demonstrate the presence of a fistula.[6]

FIG 1. Computed tomographic scan discloses a 4.0 x 2.7 x 2.3 cm abscess (white arrow) between the hypopharynx and upper part of right thyroid lobe

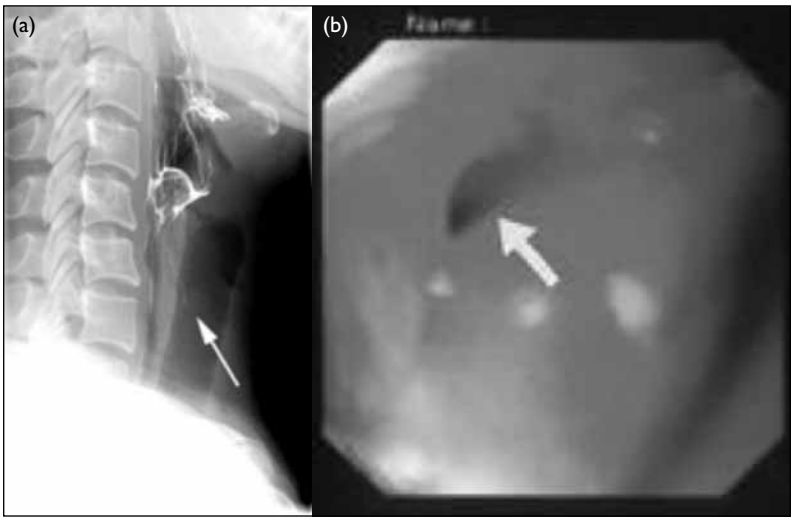

FIG 2. (a) A sinus tract of 1.8 cm (white arrow) was demonstrated to arise from the apex of right pyriform sinus. (b) Endoscopic view displaying the internal orifice (white arrow) of the fistula tract

The origin of PSF can be elusive. Miyauchi et al[1] favoured the third pharyngeal pouch theory as thymic tissue (derived from the third branchial pouch) that has been identified along the duct in some of their patients. By contrast, some clinicians embrace to the fourth pouch theory based on the anatomic finding that the tract is between the superior laryngeal nerve (fourth arch derivative) and the recurrent laryngeal nerve (sixth arch derivative).[4] More recently, the ultimobranchial body (fifth pouch) theory was proposed to be a possible mechanism for PFS pathogenesis[4,7] as calcitonin-positive cells originated from the ultimobranchial body were found near the PSF. Asymmetric (absent or restricted) development of the ultimobranchial body on the right side can explain the rarity of right PSF clinically.[7] Nevertheless, our case was more compatible with the fourth pouch mechanism as calcitonin-positive cells were absent on immunohistochemical examination and the tract traversed the inferior constrictor (fourth arch derivative).

The differential diagnoses of a PSF with abscess formation encompass an infected thyroglossal cyst, branchial cyst, or subacute thyroiditis. Infected thyroglossal cysts are usually located at the midline of the upper anterior neck at the level of hyoid bone, whereas infected branchial cysts present as lateral neck swellings, part of which is under the sternomastoid muscle. Primary suppurative infection of the thyroid is rare as the high iodine content and rich vascularity impede bacterial invasion. Subacute thyroiditis might mimic an abscess originating from PSF clinically as both manifest as painful anterior neck swellings. The CT scan findings of subacute thyroiditis will reveal mainly swollen thyroid gland. Conversely, a PSF abscess is a perithyroid swelling with its epicentre outside the thyroid gland.

Conclusion

This case illustrates a rare but clinically important problem. Clinicians should be aware that right-sided PSF can occur, and can be one of the differential diagnoses of spontaneous anterior neck abscesses on the right side. Appropriate investigation and surgical extirpation of the sinus tract, as would be undertaken for left neck counterparts, can prevent unnecessary recurrent abscess in right neck.

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