



## Pyriiform Sinus Fistula

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### ABSTRACT

Pyriiform sinus fistula is the rarest of the cervical branchial anomalies. A recurrent left sided lower neck infection may be the only clue to this elusive entity. The ambiguity of the presentation, its diagnosis, delineation of its possible anatomical course and treatment options are highlighted in this review article.

### Introduction:

Development of the neck takes place from the branchial apparatus which is an embryological complex. Abnormal development gives rise to branchial anomalies that most commonly present with neck masses and occasionally with external opening on the skin forming sinuses. Most often they are undiagnosed till multiple surgeries have been performed. Of the various branchial anomalies, the 2<sup>nd</sup> branchial anomaly is the most common. Third and 4<sup>th</sup> branchial anomalies account for only about 3 to 10%<sup>1</sup>. The latter are in close proximity or pass through the thyroid gland and originate in the pyriiform fossa, hence they are called pyriiform sinus fistula (PSF)<sup>2</sup>. Most reported cases are found on the left side with very few being reported on the right. This may be related to normal embryology of the branchial apparatus, where the fourth arch artery on the left side becomes part of aortic arch, whereas on the right side it becomes proximal part of right subclavian artery<sup>2</sup>.

### Case report:

A 19 year old male presented with complaints of recurrent swelling in the anterior aspect of left

lower neck. He had undergone multiple incision and drainage of the abscess and had received anti-tuberculosis medications and various antibiotics empirically. However, this therapy only resulted in temporary relief. On presentation to us, he had tenderness and mild swelling in the left lower neck with scars of previous procedures. Barium esophagram (BO) suggested the presence of a pyriiform sinus (Figures 1,2).

Computed tomogram (CT) showed a small tract containing air pockets and a streak of contrast, located anterior to the left carotid artery, leading to apex of the left pyriiform sinus (Figure 3). He was reluctant to try non-invasive treatment and opted to undergo surgery. He successfully underwent excision of the tract along with a left hemithyroidectomy as the tract was going through the thyroid gland. He made an uneventful recovery and has not had any recurrence for the last 6 years.

### Discussion:

PSFs occasionally present as a discharging sinus along the anterior part of sternomastoid, often with localised swelling and recurrent infection. The more typical presentation is with a recurrent neck abscess with a history of repeated surgical drainages, resulting in fistula formation. If a PSF courses through thyroid gland it may also present with acute suppuratives thyroiditis<sup>3,4</sup>.

Knowledge of the embryological relationships of the branchial clefts to the adjoining structures, can predict a probable course of 3<sup>rd</sup> and 4<sup>th</sup> branchial fistulas. A typical course of the 3<sup>rd</sup> branchial fistula

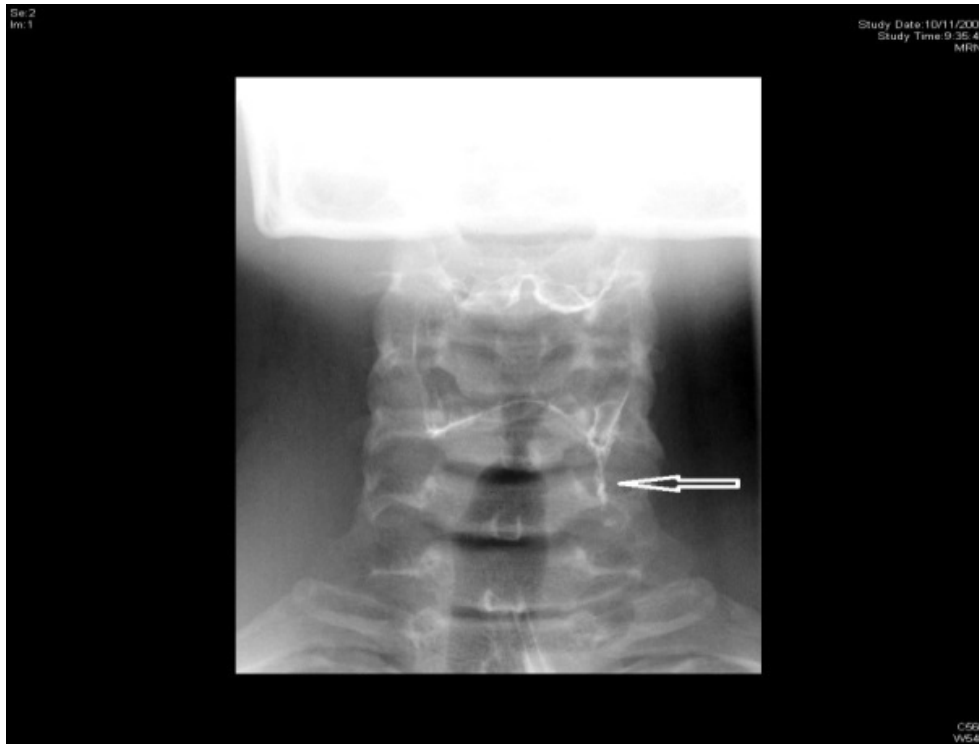


Figure 1: Barium esophagogram AP view showing tract delineated by barium. (white arrow)

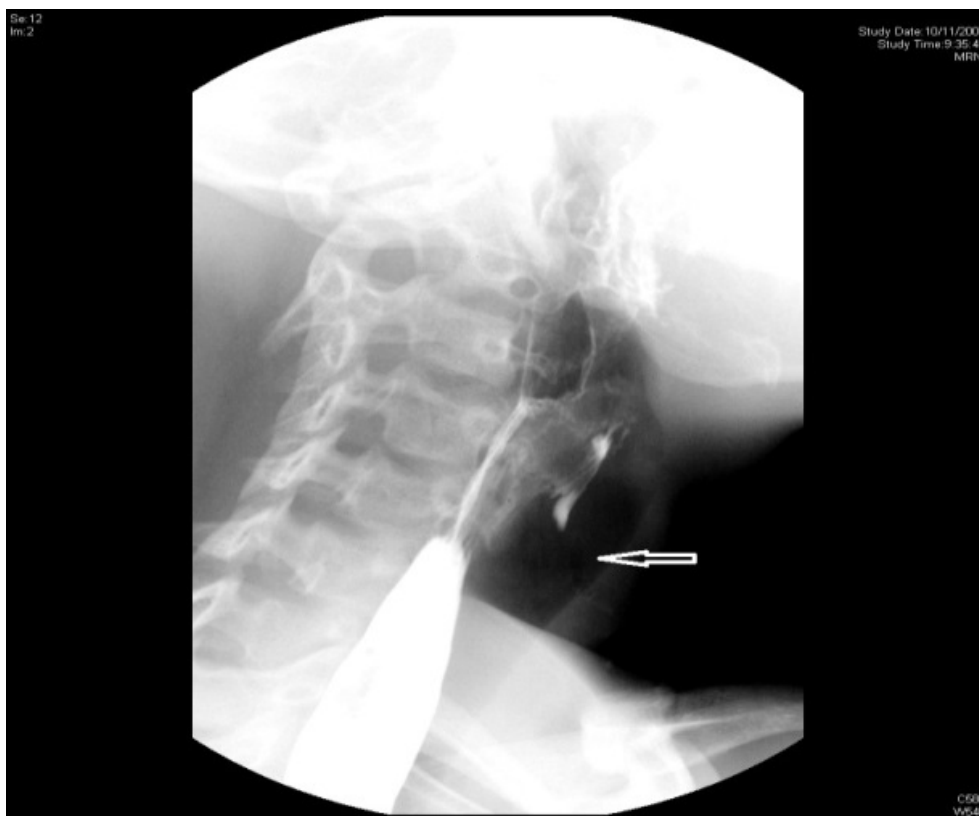


Figure 2: Barium esophagogram LAT view proximal showing barium filled tract with 'air pockets' in the distal part, leading to skin surface. (White arrow)

is to pierce the platysma, ascend along carotid sheath, pass over superior laryngeal nerve, deep to glossopharyngeal nerve (4<sup>th</sup> arch nerve), pass behind the internal carotid artery, pierce the thyrohyoid

membrane and enter upper lateral pyriform sinus<sup>5</sup>. The expected course of a 4<sup>th</sup> branchial fistula would also pierce platysma; ascend along carotid sheath but pass under superior laryngeal nerve and over

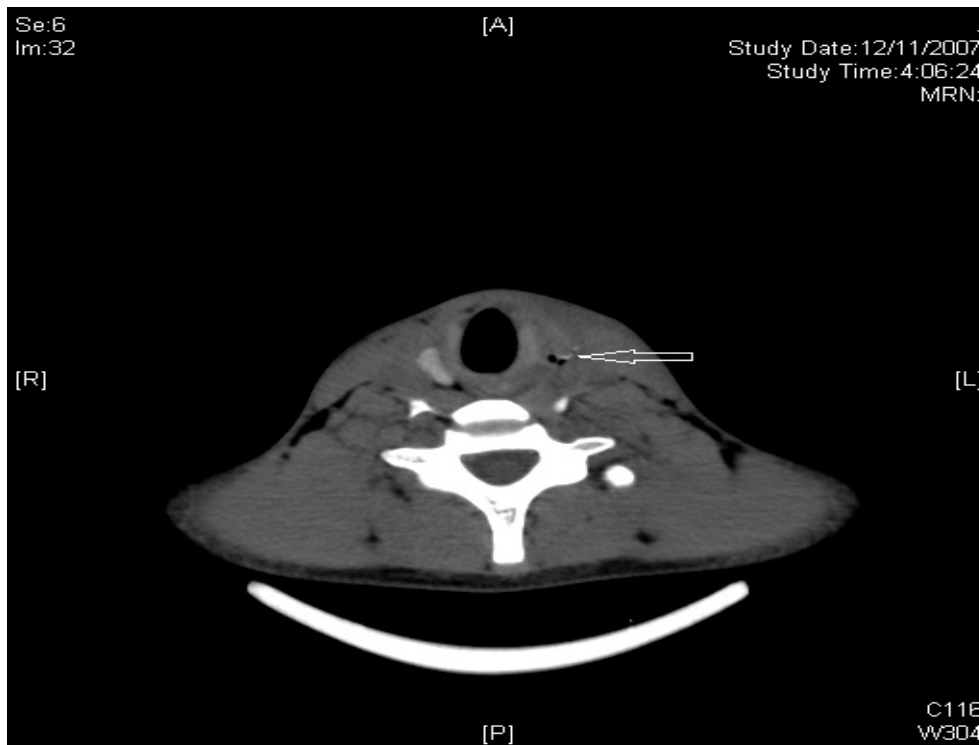


Figure 3: CT scan axial image showing 'air-pockets' with streaks of contrast near apex of left pyriform sinus. (White arrow)

the recurrent laryngeal and hypoglossal nerve. Then it would dip back into mediastinum, passing the aortic arch on the left and the subclavian artery on the right side. Finally, the fistula would ascend to enter larynx near cricothyroid joint or lower part of thyroid cartilage, pass through inferior constrictor muscle and enter apex of pyriform sinus<sup>4</sup>. Other differentiating features may include presence of thymic tissue in the third sinus and thyroid tissue in the fourth sinus. Although the above description is of interest, there is considerable overlap between the two. None of the PSF has been reported to follow the above course entirely. Fibrosis developing after infection makes correct identification of anatomical relationships difficult.

An alternative embryological process is perhaps pertinent to the clinical presentation of branchial cleft fistulas. As the thymus descends during fetal development, a thymo-pharyngeal duct is formed. The thymus is derived from ventral portion of third pouch and it descends through fourth arch during 7<sup>th</sup> to 8<sup>th</sup> week of intrauterine life. It fuses with its counterpart in the midline forming a single organ. The thymopharyngeal duct soon gets obliterated. Failure of this duct to close results in the formation of a branchial sinus lined by endodermal cells. These cells arise from the pyriform fossa and pass in close association to the thyroid gland as they head towards cervical inlet. This is analogous to the formation of a

thyroglossal cyst between tongue base and thyroid gland<sup>6,7</sup>.

Clinically a non-communicating cyst or communicating non infected cyst may present as a cold thyroid nodule or can be confused with a thyroglossal cyst<sup>8</sup>. The most common presentation is that of recurrent abscess with a repeated history of incision and drainage. A history of recurrent upper respiratory tract infections, neck or thyroid pain and tenderness as well as a neck mass is common. Other manifestations include cellulitis, hoarseness, odynophagia, thyroiditis, abscess and stridor.

Diagnosis of a PSF is based on demonstration of a sinus or a fistula, originating from pyriform sinus. BO has been widely used to demonstrate the sinus tract<sup>9</sup>. If there is acute inflammation, the chance of a false negative result increases due to the tract obliteration by inflammatory oedema<sup>10</sup>. Real time ultrasound can also be used to establish the connection of an abscess cavity to the pyriform sinus by performing the 'trumpet, manoeuvre (exhale with pursed lips to distend the pyriform sinus)<sup>11</sup>. A CT scan is considered the investigation of choice, since it can delineate the location and extent of a PSF. If the scan is performed soon after BO, the sensitivity is considerably increased<sup>12</sup>. Carbonated beverages have also been used as alternative to barium to demonstrate air in the fistula's tract<sup>13</sup>. In addition, the Trumpet manoeuvre can be used to

facilitate demonstration of sinus tract during BO and CT. Indirect laryngoscopy often shows the internal opening of the tract. Catheterization of the internal opening with small Fogarty embolectomy catheters may facilitate dissection. Some authors have used methylene blue dye to delineate the entire tract but extravasation can hinder identification of important structures around the tract during surgery<sup>14</sup>.

Meticulous dissection to identify superior and recurrent laryngeal nerve should be taken. The tract should be traced and dissected as high as possible towards pyriform fossa, ligated and excised. An oblique thyrotomy is performed above the cricothyroid joint to expose the apex of the pyriform sinus, thus, preventing injury to recurrent laryngeal nerve. Recurrence of the fistula can occur if the thyroid lobe is not removed with the tract, if the tract passes through the thyroid or if the resected specimen shows an absence of the epithelial lined tract. However, recurrent infection can destroy the epithelial lining. Chemo cauterization of the internal opening using trichloroacetic acid has been described with short term symptom free follow up<sup>15</sup>. Using

a laser to obliterate the epithelium has also been advocated in the treatment of PSF. An endoscopic approach with use of a carbon dioxide laser has been reported to produce good outcomes<sup>16,17</sup>. These less invasive treatments are an attractive option, since they have a shorter surgical time and a low morbidity and can be performed as an outpatient procedure.

#### Conclusion:

PSFs are uncommon developmental abnormalities of the branchial apparatus which are often misdiagnosed, resulting in multiple operations prior to making the correct. A barium esophagram often reveals the presence of a tract communicating with the pyriform sinus. However, a CT scan is considered as the investigation of choice. Complete excision of the tract, including its entry into the pyriform sinus with or without hemithyroidectomy, is essential to prevent recurrence. However, there have also been an increasing number of case reports managed by less invasive procedures, such as chemo or electro-cauterization that have obtained good clinical outcome and reduced morbidity. More research in this area is needed.

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