

Pyriform Sinus Fistula

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Three patients had cervical draining sinus communicating with the pyriform sinus and one patient had acute suppurative thyroiditis resulting from infection through the pyriform sinus. There was a moderate to severe perithyroidal inflammation in all 4 cases. The age of onset ranged from 7 to 18 years old (mean; 12) but that of confirmation 9, 15, 18 and 67 years of age. Three of the patients were male and 3 of the cases involved the left side. All patients had suffered from several recurrences of cervical abscess, ranged from 2 to 7 times (mean; 4). Characteristic clinical features included 1) onset at a young age 2) frequent recurrence unless the fistula was extirpated completely 3) presenting with cervical draining sinus after repeated incision and drainage. It is supposed that the fistula is a route of infection mainly in the perithyroidal space and subsequently into the thyroid gland. When the fistula communicates directly with the thyroid gland, it can cause primary acute suppurative thyroiditis. Chronic cervical draining sinus with histories of repeated incision and drainage may be the clue to the diagnosis. A barium paste swallow study is the radiologic procedure of choice and complete removal of the fistula is the treatment of choice.

Key Words: Pyriform sinus fistula, acute suppurative thyroiditis, cervical draining sinus

Since the thyroid gland is very resistant to bacterial infection and is not in contact with the outside, acute inflammation is unlikely to occur unless there is some underlying abnormalities. There are several possible routes of infection in acute suppurative thyroiditis, such as septicemia (Hawbaker, 1971) and pyriform sinus fistula (English and Al-Hussani, 1983; Har-Elr *et al.* 1991). The pyriform sinus fistula ends in or adjacent to the thyroid and allows the development of bacterial infection in or around the gland (Miyauichi *et al.* 1981).

We experienced 4 cases of pyriform sinus fistula. Except for one case, three were communicated with a previous incisional scar and those caused chronic cervical draining sinus rather than acute suppurative thyroiditis. The ipsilat-

eral thyroid gland was relatively intact except for a marked pericapsular inflammation.

This report reviews and discusses the symptomatology, embryology and problems involved in the surgical management of pyriform sinus fistula.

CASE REPORTS

Case I

A 67 year-old man developed a draining sinus on the left anterior lower neck. His medical history was remarkable for several episodes of recurrent anterior neck abscess in the same location since 7 years of age. These were treated with incision and drainage and antibiotics. Some purulent discharge was noted at the same site since 60 years of age and recently it has been aggravated. A careful physical examination revealed a pin-point external opening at the same location. A barium paste swallow study (Fig. 1) showed a tract extending from the apex of the left pyriform sinus. A sinogram

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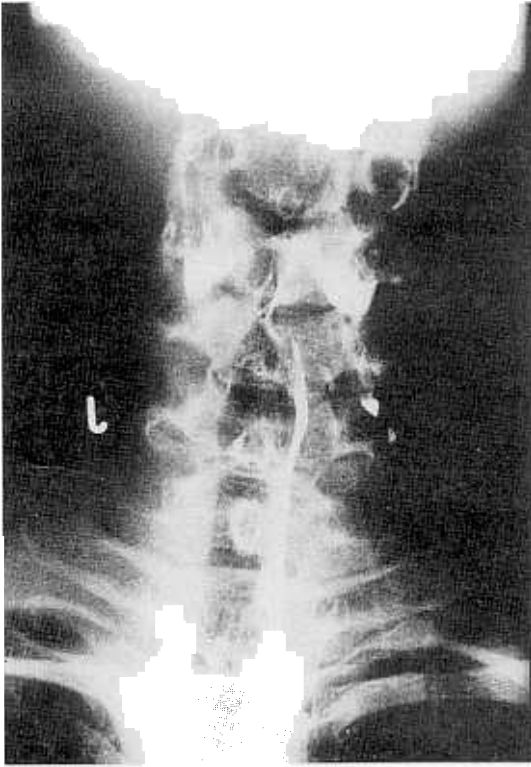


Fig. 1. Esophagogram demonstrates the fistula extending from the apex of the left pyriform sinus to the left anterior neck (case I).

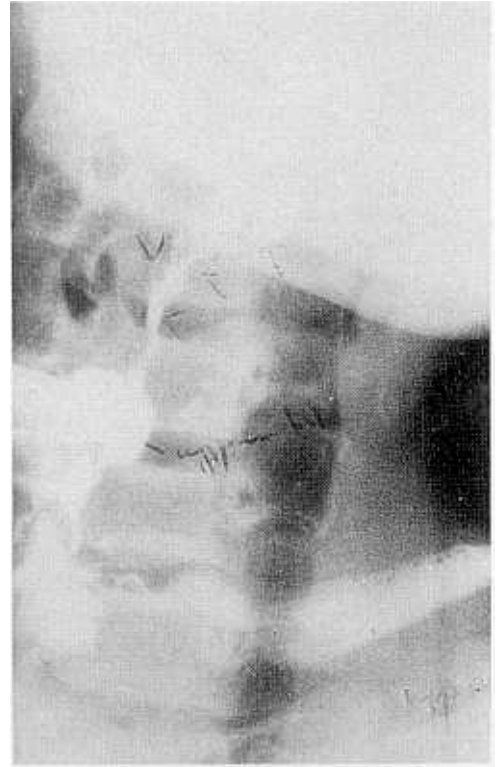


Fig. 2. Sinogram shows a fistulous tract extending from the right anterior neck to the apex of the right pyriform sinus (case II).

also showed a fistulous tract extending from the external cutaneous orifice to the left pyriform sinus. Exploration revealed a $2 \times 2 \times 3$ cm sized inflammatory mass attached to the posterolateral aspect of the midportion of the left thyroid gland. And a well formed fistulous tract was along the anterior border of the sternocleidomastoid muscle to the left pyriform sinus. Partial thyroidectomy and complete removal of the fistulous tract and debridement were performed. Seven and one half years after the operation, the patient shows no evidence of residual or recurrent disease.

Case II

A 23 year-old man was admitted because of a recurrent draining sinus on the right anterior neck. His medical history was remarkable for three episodes of recurrent neck abscess in the

same location since 18 years of age. These were treated with incision and drainage or excision and debridement with a good response. A sinogram (Fig. 2) showed a fistulous tract extending to the right pyriform sinus and a barium paste swallow study revealed a tract extending from the right pyriform sinus. Exploration revealed a well formed fistulous tract extending to the right pyriform sinus. Complete excision of the tract and debridement without thyroidectomy were performed. Seven and one half years after the operation, the patient is well without evidence of residual or recurrent disease.

Case III

A 29 year-old woman presented with a painful and tender left anterior cervical mass. Her medical history was remarkable for two epi-

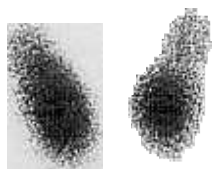
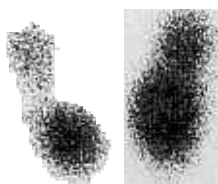


Fig. 3. Thyroid scan shows decreased uptake of the upper part of the left lobe of the thyroid gland (case IV).



sodes - 15 years-old and 18 years-old - of recurrent anterior neck abscess. These were treated with incision and drainage and antibiotics. Thyroid function tests were normal and a thyroid scan showed a cold defect on the upper pole of the thyroid gland. Neck ultrasonography revealed an ill-defined mixed echogenic mass between the carotid artery and left thyroid gland. Needle aspiration showed marked inflammatory cells without malignancy. Exploration revealed an inflammatory mass measuring about $2 \times 2 \times 2$ cm, which was attached to the mid-lateral portion of the left thyroid gland. A thin fibrous tract ran cephalad along the sternocleidomastoid muscle but disappeared near the upper pole of the thyroid gland. Left lobe of the thyroid gland and the inflammatory mass were



Fig. 4. CT scan demonstrates an ill-defined low-density mass (cyst or abscess) in the left thyroid gland with perithyroidal inflammatory changes (case IV).

removed. On the 4th postoperative day, the lower collar operative incision seemed to be infected and a pus-like discharge was noted. A conservative management was useless, so reexploration was carried out on the 11th postoperative day. Reexploration revealed a well formed fistulous tract extending from the operative wound to the left piriform sinus. Completed excision of the fistulous tract and debridement were performed. Two years and 4 months after the operation, the patient shows no evidence of residual or recurrent disease.

Case IV

A 9 year-old boy was transferred due to the recurrent tender left anterior neck mass. He was well until 3 months before his first attack of the tender anterior neck mass, which was resolved with intravenous antibiotics at a private clinic. About 1 month before, the present mass reappeared and it did not subside with antibiotics therapy. So he was referred to our hospital. On physical examination, there was a 4×5 cm sized ill-defined, diffuse, tender mass on the left anterior neck. A thyroid scan (Fig. 3) showed a cold defect on the left upper pole. C-T scan (Fig. 4) revealed a cyst/abscess in the upper pole of the left thyroid gland and left perithyroidal space. The patient was euthyroid, both clinically and chemically. Exploration was

performed. The upper pole of the left thyroid gland was upwardly retracted and a fibrous tract between the thyroid and the left pyriform sinus was noted. Left lobectomy and debridement were performed. The patient did well postoperatively and, 6 months later, shows no evidence of recurrent or persistent disease.

DISCUSSION

Since the thyroid gland is very resistant to bacterial infection, acute suppurative inflammation is rare unless there is some underlying abnormality. In recent years, there have been increasing reports of acute thyroiditis or perithyroidal inflammation caused by the pyriform sinus fistula. The fistula originates from the apex of the pyriform sinus, penetrates the cricopharyngeus or cricothyroid muscle, runs anteriorly, and then ends next to or enters a lobe of the thyroid (Miyauchi *et al.* 1990; Miyauchi *et al.* 1981) (case IV) or connected to the incisional scar as a draining sinus (case I, II, III).

There is some debate about the origin of the lesion. There was some clinical and histologic evidence that supports the theory that the third branchial pouch origin is the presence of thymic tissue in their inflammatory mass or in the cyst wall (Burge and Middlestone, 1983; Miller *et al.* 1983). Many investigators found the tract to course cephalad to the recurrent laryngeal nerve, which is a sixth branchial arch nerve, and caudal to the superior laryngeal nerve, which is a fourth branchial nerve (DeLozier and Sofferman, 1986; Sandborn and Ahafer, 1972; Tayer *et al.* 1982; Tovi *et al.* 1985; Tucker and Skolnick, 1973). Only a tract of fourth pouch origin can run this course (Hae-El *et al.* 1991).

The abscess, resulting from the pyriform sinus fistula, seems to be formed mainly in the perithyroidal space (case I, II, III) but is represented as acute suppurative thyroiditis (case IV) in the case of direct communication with a lobe of the thyroid. A fistula between the incisional scar and the pyriform sinus may eventually develop after incision and drainage (case I, II) or incomplete removal of the fistula (case III). Bacteria from the fistula seem to spread along the perithyroidal space around the

thyroid and invades the thyroid gland secondarily (Komada *et al.* 1987; Takai *et al.* 1979). Inflammation may occur in the thyroid in a case with a fistula entering a lobe of the thyroid (Miyauchi *et al.* 1981). Hirata *et al.* (1984) and Miller *et al.* (1983) have reported cases of an intrathyroid cyst with fistulous communication to the pyriform sinus. Since the thyroid gland is actually involved whether primarily or secondarily, the pyriform sinus fistula should be recognized as the most common underlying abnormality in acute suppurative thyroiditis (Miyauchi *et al.* 1990).

Takai *et al.* (1979) recommended a careful search of the hypopharynx for the fistula in all patients with acute suppurative thyroiditis of unknown origin, especially in young patients with recurrent abscess. Har-El *et al.* (1991) suggested that a patient who develops acute suppurative thyroiditis and/or thyroid abscess for the first time should have the appropriate work-up to try to document internal pharyngeal sinus. The barium paste swallow study and computed tomographic scan are the radiologic procedures of choice (Takai *et al.* 1979). Patients with noninfected cystic thyroid masses, especially if on the left side, should have the same work-up (Har-El *et al.* 1991). The patient with recurrent thyroid abscess or perithyroidal abscess should have the same work-up.

Incision and drainage and intravenous antibiotics are not the definitive treatment of recurrent perithyroidal abscess with a documented or suspected branchial etiology. After confirmation, early excision of the cyst and tract with partial or total ipsilateral thyroid lobectomy minimizes morbidity. The tract should be double ligated and divided at the pyriform sinus. If exposure is extremely difficult, resection of the superoposterior segment of the thyroid ala will bring the pyriform sinus into direct view and will facilitate dissection (Har-El *et al.* 1991). English and Al-Hussani (1983), Miyauchi *et al.* (1981) and Takai *et al.* (1979) emphasized that complete removal of the fistula was essential for a permanent cure because recurrence is very common. Miyauchi *et al.* (1990) reported that about 62% of the patients who declined fistulectomy have not had a recurrence so far, and none of the patients who had a complete fistulectomy developed a recurrence, confirming the effectiveness of surgery. So they suggested that a spontaneous cure may be pos-

sible in some patients especially those with a very fine fistula. A follow-up policy without fistulectomy in patients with a very fine fistula and no previous episodes may be acceptable (Miyauchi *et al.* 1990). We believe that a complete excision of the fistula is essential for cure. And removal of the ipsilateral thyroid gland is necessary in some cases but not mandatory.

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