

A case report: Fourth branchial cleft fistula successfully treated with microlaryngoscopic cauterization

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Abstract

A 23-year-old man was presented with recurrent left-sided neck abscess which is characteristic clinical features of fourth branchial cleft fistula. The abscess burst spontaneously, forming a fistula. Diagnosis of fourth branchial fistula in this patient was verified by barium radiograph and injection of methylene blue into the external opening. An internal opening was found at the apex of the left piriform fossa. The current treatment of choice involves complete excision of the sinus tract. Endoscopic cauterization of the internal fistulous tract was developed by some authors. It offers several advantages over open excision such as less injuries to important structures. The cost and duration of treatment are also reduced. Microlaryngoscopic electric cauterization of the internal opening was used to close the fistula. There has been no recurrence of the cervical abscess during 10 months of follow-up. Therefore, microlaryngoscopic electric cauterization seems to be a safe treatment for patients with fourth branchial cleft fistulas.

Key Words: branchial cleft fistula

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Fourth branchial cleft fistula ที่ประสบความสำเร็จจากการรักษาด้วยวิธี microlaryngoscopic cauterization: รายงานผู้ป่วย 1 ราย

นิรันดร์ หุ่นฉายศรี

ภาควิชาจักษุ โสต ศอ นาสิก ลาริงซ์วิทยา คณะแพทยศาสตร์ มหาวิทยาลัยศรีนครินทรวิโรฒ

บทคัดย่อ

Fourth branchial cleft fistula พบได้น้อยมาก ผู้เขียนได้รายงานผู้ป่วย 1 ราย ที่มีลักษณะเฉพาะคือเป็นฝีหนองที่คอแบบเป็นๆ หายๆ จนเกิดรูทะลุที่ผิวหนังบริเวณคอ สามารถวินิจฉัยโดยการให้กลืนสาร Barium และการฉีดสี methylene blue ผ่านรูเปิดที่คอ แล้วเห็นรูเปิดด้านในบริเวณยอดของ pyriform fossa ข้างซ้าย การรักษาจำเป็นต้องผ่าตัด sinus tract ออกให้หมด ต่อมามีการใช้วิธีจี้รูเปิดด้านในเพื่อการรักษาโรคนี้ ซึ่งพบว่าวิธีนี้ได้เปรียบกว่าวิธีการผ่าตัดแบบเปิดหลายอย่าง เช่น ลดอันตรายต่อโครงสร้างสำคัญที่คอ ลดระยะเวลาในการรักษา และช่วยลดค่าใช้จ่ายอีกด้วย ในผู้ป่วยรายนี้พบว่าการรักษาด้วยวิธีจี้รูเปิดด้านในด้วยกระแสไฟฟ้าผ่าน microlaryngoscope ได้ผลดี และยังไม่พบการเกิดฝีหนองซ้ำในช่วงเวลาติดตาม 10 เดือน ดังนั้นการรักษาด้วยวิธีจี้ด้วยกระแสไฟฟ้าผ่าน microlaryngoscope น่าจะเป็นการรักษาที่ปลอดภัยสำหรับผู้ป่วย fourth branchial cleft fistula

คำสำคัญ: branchial cleft fistula

Branchial cleft or pouch anomalies (sinuses, cysts and fistulas) arise from abnormal development of the embryological complex known as the branchial apparatus. These anomalies most commonly present with a cervical mass or cyst, or with drainage from a cervical sinus opening. They account for up to 17% of pediatric neck masses¹. Third and fourth branchial pouch sinuses, however, are quite rare, accounting for only 3-10% of all branchial anomalies^{2,3}. These abnormalities often occur as tracts from the piriform sinus to the thyroid gland and are sometimes referred to as piriform sinus fistulas. Third and fourth branchial fistulas usually present as fluctuant neck masses located at the anterior border of the sternocleidomastoid muscle. These branchial fistulas may also be associated with neck abscess or acute suppurative thyroiditis⁴. Treatment has traditionally involved excision of the sinus tract with partial thyroidectomy, if the thyroid gland is involved, Cannulation of the piriform sinus fistula during direct laryngoscopy to aid complete surgical excision has also been described in literature⁵. Some reports

have described endoscopic cauterization of fourth branchial cleft sinuses with good results^{6,7}. This article reports an additional case of fourth branchial cleft fistula which is effectively treated with microlaryngoscopic electric cauterization.

Case Report

A 23 year-old man presented with an abscess on the left side of neck. He had the history of neck abscess since childhood, but there was no recurrence for more than ten years. Last 2 years, he started to have few episodes of neck abscess. He was treated at private hospital with medication and surgical drainage. After the last episode, an opening was seen at the prior abscess area. There was frank leak from the fistula on oral intake of liquids. He was referred to HRH Princess Maha Chakri Sirindhorn Medical Center for further management. On examination, there was a pinhead-size opening on the left side of neck, just below the cricoid cartilage, with scarring around the opening (Fig 1). Barium swallow showed spillage of the dye in the tract from left piriform sinus area to skin (Fig 2).



Figure 1 Suction tip pointed at the opening of the fistula.



Figure 2 Barium swallow showed fistulous tract from piriform sinus to skin (arrow).

He was scheduled for microlaryngoscopic cauterization under general anesthesia. At surgery, after suspension microlaryngoscope was applied, the internal opening of the fistula was identified by injection of methylene blue into the external opening. The dye was seen coming from the apex of the left piriform sinus, anteriorly. The size of the internal opening was approximately 2 mm. in diameter. The opening in piriform sinus was cauterized by suction cautery until it became blanching. He was admitted for 3 days for observation. Nasogastric tube feeding was advised for 2 weeks postoperatively. The external opening was closed at 1-week follow-up period. Lastly, he had no recurrence of neck abscess during ten months postoperative period.

Discussion

The human branchial apparatus comprises 5 paired mesodermal arches, separated by 4 pairs of endodermal and ectodermal invaginations that are called pouches and clefts. A branchial fistula forms when there is remnant of both the

the pouch and the cleft with rupture of the interposed branchial plate. A sinus is formed when the tract opens either to the gut or to the skin but not both, whereas a cyst is formed when there is no communication to both of them⁸. It has been found that 95% of the congenital anomalies of the branchial apparatus are derived from the second branchial arch, pouch, and cleft. The remaining anomalies stem from the first and third arch. The remnants of the fourth branchial arch are very rare⁹⁻¹⁰. The theoretical course of a third branchial fistula would pass deep to third arch structures and superficial to fourth arch structures. More specifically, it would arise at the skin near the mid-to-lower third of the anterior border of the sternocleidomastoid muscle. A third branchial pouch fistula would pierce the platysma, ascend along the carotid sheath, pass over the superior laryngeal nerve and deep to the glossopharyngeal nerve, pass behind the internal carotid, pierce the thyrohyoid membrane, and enter the upper lateral piriform sinus wall¹¹.

The theoretical course of a fourth branchial fistula would originate at the anterior border of the lower portion of the sternocleidomastoid muscle. It would also pierce the platysma, ascend along the carotid sheath, pass under the superior laryngeal nerve, but over the recurrent laryngeal and hypoglossal nerves. A fourth branchial pouch sinus would then dip back into the chest to pass around the aortic arch on the left and the subclavian artery on the right. Finally, it would ascend to enter the larynx near the cricothyroid joint or through the lower horn of the thyroid cartilage, pass through the inferior constrictor, and enter the apex of the piriform sinus¹²⁻¹³. It is probably due to the complex and convoluted course that a complete fourth branchial fistula has yet to be reported. Fourth branchial pouch anomalies typically present as a sinus or fistula from the apex of the piriform sinus to the deep side of the thyroid gland¹². Approximately, 83-97% of them are on the left side¹⁴⁻¹⁵. It has been suggested that this is due to asymmetrical vascular development¹⁶.

Because of the similarity in their course, it is difficult to differentiate between the fourth and the third branchial arch anomalies. Both sinuses are expected to open into the piriform sinus. To differentiate between these anomalies, surgical exploration is required. If the sinus passes below the superior laryngeal nerve, a fourth branchial pouch sinus is suggested. On the other hand, if the sinus passes above the superior laryngeal nerve, a third branchial pouch sinus is suggested¹⁷. Another clue to the origin of the anomalies is the location of the sinus opening in the piriform sinus seen from endoscope. An opening in the apex of the piriform sinus suggests fourth branchial cleft anomalies⁶.

Regardless of the pouch of origin, histologic examination of excised fistulous tracts often reveals inflammatory changes to nonkeratinized stratified squamous or stratified columnar epithelium. Transitional epithelium and subepithelial lymphoid infiltrates have also been reported¹⁸. While controversy remains regarding identification of third and fourth branchial sinuses, the current treatment of choice involves complete excision of the sinus tract. Some authors develop new treatment by electric cauterization^{6,7} and chemocauterization^{19,20} of the internal sinus or fistulous tract for closing them. Jordan et al⁶ reported a series of seven patients who underwent endoscopic cauterization of the sinus opening, four of them had no evidence of recurrence during 18 months of follow-up. Verret et al⁷ reported a retrospective study to evaluate the effectiveness of endoscopic cauterization as definitive treatment for fourth branchial cleft sinuses. Seven of the 10 patients treated with endoscopic cauterization of the fourth branchial cleft sinuses showed no recurrence with an average follow-up of 3 years. Three of the patients were unavailable for follow-up, but medical records of the hospital showed no additional admissions for those patients for neck masses. No morbidity of the procedure was identified.

The patient in this article was a case of fourth branchial arch fistula, which manifested in childhood with typical history of left-sided neck abscess. The abscess was recurrent and finally formed a complete fistula, communicating from neck to the apex of left piriform fossa. The diagnosis was confirmed by a contrast study (barium swallow) and injection of methylene blue into the fistula. Under direct laryngoscope, dye was seen coming from the internal opening at

the apex of piriform sinus. Endoscopic cauterization offers several advantages over the open procedure. Because of previous infection and scar tissue, difficult dissections can be avoided by endoscopy. As no open excision is required, the risk of injuries to important structures are eliminated, i.e. injuries to the superior and recurrent laryngeal nerves, esophagus, trachea, and aorta. Also, the duration of treatment and the cost are reduced in endoscopy. Microlaryngoscopic cauterization was used in this patient because of the above reasons. Despite no recurrence of neck abscess during ten months postoperative period, long term follow-up is needed.

Conclusions

The diagnosis and management of fourth branchial pouch anomalies are challenging. Diagnosis requires a high index of suspicion, from the clinical presentation of recurrent neck or thyroid abscess. Barium radiography and injection of methylene blue are useful to confirm the presence of a fistulous tract opening into piriform apex. Microlaryngoscopic electric cauterization seems to be an effective primary treatment for patients with a piriform sinus fistula.

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