Congenital Sialo-Cutaneous Fistula of the Parotid Salivary Gland: A First Branchial Cleft Anomaly: A Case Report

La fistule sialo-cutanée congénitale de la parotide salivaire: une première anomalie branchiale fendue: un rapport de cas

Da Lilly-Tariah, O. B.
Department of Surgery, College of Health Sciences, University of Port Harcourt, Port Harcourt

Abstract

Anomalies in the development of the branchial apparatus may result in the formation of cysts, sinuses and fistulas in the head and neck region. These anomalies are rare and when they do occur, the first branchial cleft fistulas are even rarer with less than 50 cases reported worldwide. A case of first branchial cleft anomaly presenting as a congenital sialo-cutaneous fistula is hereby reported.

Key Words: branchial, cleft, sialo-cutaneous, fistula.

Resume

Les anomalies dans le développement de l'appareil branchial peuvent aboutir à la formation de kystes, de sinus et de fistules dans la région de la tête et du cou. Ces anomalies sont rares et quand elles surviennent, les premières fistules branchiales fendues sont encore plus rares puisqu'il y en a moins de 50 cas rapportés mondialement. Nous rapportons ici le cas d'une première anomalie branchiale fendue présentée comme une fistule sialo cutanée congénitale. ***Mots-clés:*** branchial, fendu, sialo - cutané, fistule.

Introduction

The branchial apparatus develops in human beings between the third and fourth week of embryonic life[1-3]. Developmental anomalies may affect the branchial apparatus including the first branchial cleft. The anomalies of the branchial cleft may present as cysts, sinuses or fistulas[1-6]. The anomalies are thought to arise because of incomplete obliteration of the branchial cleft[1].

Anomalies of the first branchial cleft account for about 8% of total branchial cleft anomalies[2,5] with less than 50 cases reported worldwide[2]. Of these, cysts are known to occur twice as much as fistulas or sinuses[5]. These anomalies may present in the middle ear, nasopharynx and down to below the mandible[2]. The site of the fistula, sinus or cyst may be variable[2,3].
In this report, a first cleft anomaly opens between the parotid salivary gland duct opening and in the neck, below the mandible is reported.

Case Report

A 15-year-old boy presented at the out patient unit of ear, nose and throat (ENT) department with complaints of a hole that was discharging clear liquid from the right side of the neck since birth. The discharge was more copious while eating and drinking, but unaffected by swallowing. There was no associated pain, swelling, fever or cough. His past medical history was not contributory. He had not sought surgical attention for his problem before now. He had no other birth defect. He was the first of five



Fig. 1: Cutaneous opening of the fistula.

Correspondence: Da Lilly-Tariah, O.B., Dept. of Surgery, Coll. of Health Sciences, University of Port Harcourt, Port Harcourt.

Copyright © 2006 African Journal of Paediatric Surgery
Published by Child Health Ventures



Fig. 2: Dye tracking out of the Stenson's duct

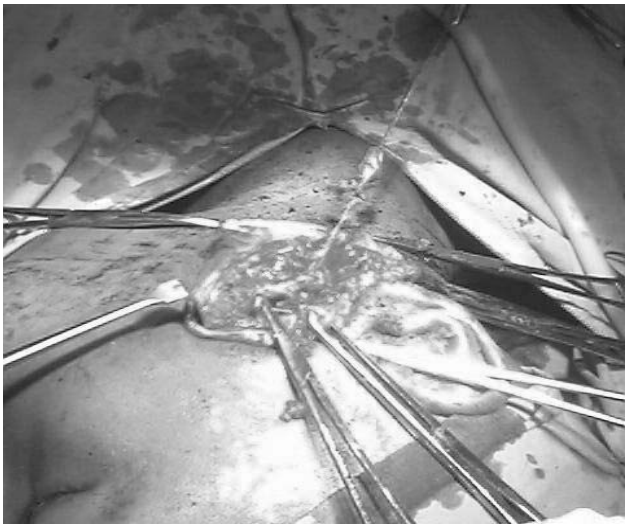


Fig. 3: Fistula tract dissected out through superficial parotidectomy.

children, and there was no family history of this problem or any other birth defect.

Physical examination revealed a healthy-looking boy. His temperature was 36.5^{0}C, pulse rate of 70 beats per minute, blood pressure of 110/70mmHg and respiratory rate of 25 cycles per minute.

In the anterior triangle of the neck, there was a pinpoint opening at the anterior border of the sternomastoid muscle on the right side at the level of the upper third of the muscle. There was a clear and slimy liquid that was discharging from the opening (fig. 1). There was no tenderness, swelling or bleeding around the opening. The neck did not reveal any other abnormality. The opening did not move with swallowing or protrusion of the tongue. No abnormality was detected on examination of the ear, nose and throat.

Ultra sound scan of the neck showed normal thyroid gland in the normal position. A sinogram using barium sulphate showed a short sinus tract that ended in the neck.

Injection of gentian violet solution through the sinus opening did not produce a back flow. Alarmed at where the dye was tracking, an examination under anaesthesia was done and the dye was seen tracking out of the Stenson's duct opening (fig. 2).

Surgery involved the use of an elliptical incision to isolate the cutaneous opening of the fistula. Through an S-shaped skin incision, a superficial parotidectomy was done. The fistulous tract was traced to the main trunk of the parotid salivary gland duct (fig. 3). The duct was ligated and excised with the fistulous tract and the salivary gland. The wound was closed in two layers with a closed suction drain in-situ. The drain was removed after 5 days. Stitches were removed on the 7th day post surgery. The wound site drained saliva till 15 days after surgery.

Histology of the specimen revealed stratified squamous epithelium. The patient has remained free of any symptoms for over 12 months of follow up.

Discussion

Congenital sialo-cutaneous fistula may present later in life with a mean age at presentation of 18.9 years[3,7]. On occasion an infected cyst ruptures and gives rise to a fistula or sinus, but the absence of pain and swelling in this report ruled out this possibility. The late presentation in this report is thought to be due to poverty and ignorance, because the discharge was described as consistently clear and slimy pointing to its salivary nature. Although otorrhoea is the most common symptom of first cleft fistulas[2,3,8], the predominant feature in this case was discharge of saliva in the neck.

First branchial cleft anomalies have been classified in different ways, based on its varied presentations. Anatomical classification by Arnot[3,9] designated type I

Copyright © 2006 African Journal of Paediatric Surgery
Published by Child Health Ventures

anomaly as defects in the parotid region, appearing in early or middle age. Type II defects occur in the anterior cervical triangle and communicate with the external auditory canal; they are typically seen in childhood. Work[2,3,10] proposed a histological classification in which type I anomaly is a cyst that is ectodermal in origin and is a duplication of the membranous external auditory canal. Type II anomaly may be a cyst, sinus or fistula and is both ectodermal and mesodermal in origin (it may contain skin, cartilage etc) and is a duplication of the external auditory canal. The classification by Olsen et al had more bearing with diagnosis and surgical management[11,12,13]. In this classification the anomalies may be cysts, sinuses or fistulas. Based on the presentation and histology report, the present defect is classified as Work type [12].

The work up of the patient in this report was based on clinical evaluation and contrast study only because of unavailability of computerized tomography in the centre. Contrast sinogram gave an impression of a sinus, which was confirmed by intra-operative dye injection of the fistula[3]. These investigations guided the surgical management and outcome in this patient. The failure of the contrast to outline the fistula may have been due to faulty technique.

The objective of treatment is to completely remove the fistulous tract while preserving the facial nerve[2,3]. This was achieved through superficial parotidec-tomy and excision of the Stenson's duct.

In conclusion, the first branchial cleft anomalies may be seen in areas in the Pochet's triangle without involving the external auditory canal. Classifications along anatomical and histological lines are not helpful in the diagnosis.

References

1. Golledge J, Ellis H. The aetiology of lateral cervical (branchial) cysts: past and present theories. J. Laryngol Otol 1994; 108: 653-659.
2. Nicollas R, Paris J, Triglia J M; Congenital lateral cysts and fistulas of the neck. Http://www.bapo.org.uk/8th_ESPO/data/htm/2725.htm. Assessed 14/04/2005
3. Tham Y S, Low W K; First branchial cleft anomalies have relevance in otology and more. Ann. Acad Med Singapore 2005. 34: 335-338.
4. Triglia JM, Nicollas R, Ducroz V, Koltai PJ, Garabedian EN. First branchial cleft anomalies: a study of 39 cases and a review of the literature. Arch Otolaryngol Head Neck Surg. 1998; 124: 291-295.
5. Smith J C; Neck, Cysts eMedicine 2003. http://www.emedicine.com/ENT/topic283.htm, Assessed 14/04/2005.
6. Sichel J Y, Halperin D, Dano I, Dangoor E; Clinical update on type II first branchial cleft cysts. Laryngoscope. 1998, vol 108: 1524-1527.
7. D'Souza A R, Uppal H S, De R, Zeitoun H; Updating concepts of first branchial cleft defects: a literature review. Int J Pediatr Otolaryngol 2002; 62: 103-109
8. Rushton V E, Pemberton M N; Salivary otorrhoea: a case report and review of the literature. Dentomaxillofacial Radiology. 2005; 34: 376-379.
9. Arnot R S; Defects of the first branchial cleft. S Afr J Surg 1971; 9: 93-98
10. Work W P; Newer concepts of first branchial cleft defects. Laryngoscope. 1972; 82: 1581-1593.
11. Olsen K D, Maragos N E, Weiland L H; First branchial cleft anomalies. Laryngoscope 1980; 90: 423-436.
12. Belensky W M, Medina J E; First branchial cleft anomalies. Laryngoscope 1980; 90: 28-39
13. Aronsohn R S, Batsakis J G, Rice D H, Work W P; Anomalies of the first branchial cleft. Ach Otolaryngol 1976; 102: 737-740.

Copyright © 2006 African Journal of Paediatric Surgery
Published by Child Health Ventures