TREATMENT OF PHARYNGOCUTANEOUS FISTULA ACQUIRED FROM INCISIONS AND DRAINAGE OF DEEP NECK SPACE ABSCESS IN A PATIENT WITH OCCULT THIRD BRANCHIAL ANOMALY.

BY

ORJI FT, DAHILO EA, EZEANOLUE BC, OKAFOR BC, AND OKORAFOR IJ

Department of Otolaryngology,
University of Nigeria Teaching Hospital, Enugu, Nigeria.

Correspondence:
Dr B C Ezeanolue
Department of Otolaryngology,
University of Nigeria Teaching Hospital,
Enugu, Nigeria.
e-mail: balseze@yahoo.com

SUMMARY:

AIM: *To report the treatment of an extremely rare case of pharyngocutaneous fistula that resulted from incisions of deep neck space abscess in a patient with occult 3rd branchial anomaly.*

Methodology: *clinical case report*

Case report: *OC is a 19-year old female who presented with 18-month history of a discharging opening in her anterior neck. This started after she had incisions and drainage of recurrent deep neck abscess three times at another health facility. We did surgical exploration to excise and close the fistula but only succeeded at the second attempt.*

Conclusion:

We conclude that branchial apparatus anomaly whether manifest or occult should be suspected in the aetiological factor of deep neck abscess. Caution should be exercised in incising and draining recurrent deep neck space abscess to avoid facilitating formation of fistula.

Key words: *brachial anomaly, deep neck space abscess, pharyngocutaneous fistula.*

INTRODUCTION

The occurrence of brachial cleft and pouch anomalies is rare and only a handful has been reported in the literature.

Branchial fistula occurs when there is developmental error and parts of the cleft and pouch persist with breakdown of the endoderm-mesoderm-ectoderm interface. When the tract is incomplete, a sinus with either an internal opening (remnant of a pouch) or external opening (remnant of a cleft) occurs. A brachial fistula or sinus will open anywhere on the skin of the neck along a line from the tragus down to the sternoclavicular joint on the anterior border of sternomastoid muscle. The internal opening for 2nd brachial apparatus is in the tonsillar fossa while the 3rd will

open in the piriform fossa. The 4th will open immediately below the piriform fossa[1, 2]. The external and middle ear structures, tonsils, thymus, thyroid gland, pharynx and larynx are also derived from this structure.

A reported retrospective study of patients with brachial cleft and pouch anomalies between 1948 and 1990 by G.R Ford et al, revealed a total of only 106 patients and further showed that second branchial cleft sinuses were by far the commonest anomalies accounting for eighty-four (79%) patients in their study series[3]. First branchial cleft anomalies were the next commonest accounting for 17% of the patients. Only two (1.9%) third branchial pouch anomalies were reported out of the 106 patients reviewed. Even then, diagnosis of third arch anomalies was only made at surgical operation. Only one patient presented with fourth branchial cyst[3]. A similar review by Choi et al between 1983 and 1993 revealed only 52 patients with branchial anomalies[4]. Again in their series, second branchial anomalies were the commonest accounting for 65% of the patients in their study series, followed by the first arch anomalies which accounted for 25% while third and fourth branchial anomalies accounted for 8% and 2% respectively[4].

This paper reports the treatment of an extremely rare combination of third branchial sinus that had an occult internal opening, complicated by recurrent deep neck space abscess formation and subsequently converted to pharyngocutaneous fristula by incisions and drainage procedures done for treatment of the abscesses.

CASE REPORT

OC is a 19 year old female student who presented at the Otolaryngology Clinic of University of Nigeria Teaching Hospital (UNTH) Enugu, Nigeria on the 5th August 2003. She presented with 12-year history of recurrent pain and swelling of the right side of her neck, and 5- month history of discharging opening over the same site.

Her problems dated back to 1992 at the age of 7 years when she developed right sided painful neck swelling 2 days after a local traditional healer's "*treatment procedure"* of crushing of the tonsils ("Mgbapia") for the sore throat she was then suffering from.. The swelling was treated with incision and drainage of pus in a private hospital followed with a course of antibiotic therapy. The neck swelling resolved, but recurred twice, at 2 and 9 years after the first incidence. On each occasion, it was incised and pus drained. In-between the episodes of recurrent neck swellings patient continued to experience intermittent neck cellulites at same site with a frequency of 3 – 4 times each year from the time of first incidence till this presentation. Each episode was relieved by a treatment course of antibiotics.

The skin wound, site of the last incision and drainage did not heal despite what was considered to be an adequate wound care at another private health facility. On the third month after this drainage, patient noticed that liquid feeds, water and saliva were leaking from a particular opening in the neck. She now presented to us with an opening in the right side of her neck just anterior to the mid-point of the anterior border of sternomastoid muscle surrounded by granulation tissue, and discharging mucopurulent fluid.

A working diagnosis of a branchial fistula was made and contrast fistulography requested. This revealed a smooth walled cul-de-sac about 2 cm in diameter just beneath the opening but without any definite tract outlined. During surgical exploration, a cyst with ill-defined wall was found without identifiable tract. The cyst wall and the granulation tissue were excised and wound closed, thereafter the wound healed uneventfully.

One year after this exploration, the neck swelling recurred at the same site and patient presented for the second time to our clinic. This time it was associated with severe odynophagia and fever. An incision and drainage of deep neck abscess was carried out the same day. On the fifth day after the incision and drainage, leakage of liquid feeds from the wound started again. A clinical diagnosis of pharyngocutaneous

fistula was entertained. Fistulography again could not outline a definite fistulous track.

Conservative management for eight weeks failed to achieve closure of the fistula. A second fistula excision was therefore undertaken. Preliminary rigid pharyngo-oesophagoscopy was done on the patient under general anaesthesia. During this procedure, methylene blue dye was instilled with cannula and syringe through the external anterior neck opening. The dye was observed to enter the pharynx and its internal opening identified in the piriform fossa. The fistulous track was excised from the external opening, passed medial to the common carotid artery to the internal opening in the piriform fossa. A fibrous strand extended inferiorly to the right sternoclavicular joint capsule. The track was well epithelised and easily admitted a size 10FG feeding tube. The fistulous tract was excised completely and the pharynx closed in layers. The neck wound was closed in layers with a z-plasty to the skin.

Post operatively, there was satisfactory wound healing with minimal skin scar and normal oral feeding re-established on the 14th day.

Histology report of the tissue specimen showed vascularised connective tissue, skeletal muscle and granulation tissue containing numerous foreign body type multinucleated giant cells and entrapped vegetable material from ingested food. She has since been discharged with no recurrence as at last review 15 months later.

DISCUSSION

Third branchial anomaly is a rare clinical entity and the same anomaly giving rise to pharyngocutaneous fistula is even extremely rare. Sunil et al stated that only 27 cases of third and fourth branchil arch fistulae were reported in the literature as at 1994.[5] Also, a 13 years review of 58 cases of all types of brachial remnant by Doi et al revealed that only one case had a pharyngocutaneous fistula of third brachial origin.[6] Detection of occult branchial sinus with an internal opening is difficult except as an incidental finding. Our patient most likely had an initial occult branchial sinus with an internal opening in the right piriform fossa. This seemingly symptomless sinus became subsequently infected from surrounding deep neck space abscess. The later most likely developed from spreading sepsis that occurred as a result of the crude traditional crushing of the tonsils.

Branchial fistula, sinus or cyst may be dormant or manifest when it is infected. Incision and drainage of any formed abscess may convert a sinus, with an internal opening only, into a fistula that communicates between the pharynx and the skin of the neck[2.] Most often, the internal opening is first discovered during surgical operation[2]. Preoperative sinography and fistulography may outline the tract. Treatment involves control of any existing infection of the tract and excising of the entire tract to prevent reoccurrence.

The presence of branchial apparatus anomaly was not suspected in our patient despite repeated deep neck sepsis for twelve (12) years. It was incidentally converted to a pharyngocutaneous fistula by the repeated incisions and drainage procedures done at other health facilities.

Ellis reported a 12 years old girl that had 13 operations for drainage of recurrent neck abscesses before presenting to him with a discharging opening in the neck. It was only during exploration that a fistulous tract was found which ended in the piriform fossa.[2] Similarly, Edmond et al reported the excision of three cases of third branchial anomalies as difficult. They reported that multiple attempts at excision were followed by recurrences for each of the cases due to incomplete excision of the tracts.[7]

In retrospect, we believe that the failure of the fistulography to outline patient's fistulous tract was as a result of granulation tissue and fibrosis occluding part of the fistula. In a study by Lin et al, where 16 cases of third branchial fistulas were reviewed only four (4) cases were outlined by eosophagogram preoperatively. The rest were demonstrated only at surgery.[8] The inconclusive preoperative fistulogram misled the surgeons in the unsuccessful first

exploration we did. Following the recurrence of the fistula, the tract was clearly outlined during the second exploration with methylene blue surgical dye.

CONCLUSION

The diagnosis of brachial anomaly is to be suspected in any patient with a history of recurrent deep neck abscess.

An initial preoperative pharyngo-esophagoscopy is a helpful diagnostic procedure. A fistulogram should be done to outline and visualize the tract.

Adequate surgical treatment should involve complete excision of the tract and meticulous repair of the pharyngeal wall to prevent recurrence.

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