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RECURRENT NECK INFECTION ASSOCIATED WITH FOURTH BRANCHIAL POUCH SINUS: A RARE CLINICAL ENTITY WITH DELAYED DIAGNOSIS

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ABSTRACT

Fourth branchial pouch anomalies are rare and usually present as lateral neck masses, abscesses, or acute suppurative thyroiditis. If a person has recurrent deep neck infections or abscesses, the presence of a branchial arch anomaly should be investigated. Here we discuss a case of recurrent neck abscess (left side) which was finally found out to be a case of left fourth branchial pouch sinus and various pitfalls associated with diagnosis of disease.

KEY WORDS*: branchial pouch, neck infection*

INTRODUCTION

The human branchial apparatus, which develops in early gestation, is comprised of six paired mesodermal arches, separated by endodermal and ectodermal invaginations known as pouches and clefts, respectively[1]. Each arch is composed of a mesenchymal core, which is derived primarily from neural crest cells and will ultimately develop into the skeletal and interstitial structures of the head and neck along with associated vessels and nerves. Derivatives of the fourth pouch include the laryngeal cartilages, the laryngeal and pharyngeal constrictor muscles, the superior laryngeal nerve, the left thoracic aorta, the right proximal subclavian artery, the ultimobranchial body from which the calcitonin-secreting interfollicular cells of the thyroid arise, and the superior parathyroid glands [2] .Congenital lateral cervical cysts, fistulae, and sinuses are thought to arise from the branchial apparatus. Approximately 95% of congenital anomalies of the branchial apparatus involve the second branchial arch, pouch, or cleft, while the remaining mostly arise from the first and third arches[3] . Remnants of the fourth branchial arch are extremely rare with less than 100 cases reported in the literature and account for 1–4% of all branchial anomalies[4]. These anomalies typically present as recurrent neck infections and/or abscesses or acute suppurative thyroiditis[5] .The origin and course of the anomalies are determined by the mesodermal derivatives of adjacent arches; sinus of the fourth branchial arch have a swelling in the neck on the anterior border of the lower sternocleidomastoid muscle and an internal opening in the pyriform sinus. Here we discuss a case of recurrent neck abscess (left side) which was finally found out to be a case of left fourth branchial pouch sinus and various pitfalls associated with diagnosis of disease.

Case Report: A 15-years-old male was referred to Government medical college Haldwani, Nainital for evaluation of recurrent left sided painful neck swelling since childhood. The swelling was associated with fever, neck stiffness and upper respiratory infection. He was treated by local practioners with broad spectrum intravenous antibiotics and anti-inflammatory drugs with resolution of symptoms; however there was no history of incision and drainage being done. There was no significant family history. At the time of presentation he was febrile with signs and symptoms of upper

respiratory tract infection along with painful swallowing. Neck showed a left sided swelling especially in thyroid region extending upto posterior part of neck with tenderness in cervical area (Fig No.1). Overlying skin on the swelling had erythema which later on developed fluctuation at some places. Swelling did not move with deglution. The right thyroid lobe appeared normal. There was no cervical lymphadenopathy and patient had no symptoms and signs suggestive of thyroid disease. Nose and laryngopharynx had signs of acute infection. Blood count confirmed euthyroid status of the patient. Total leukocyte count was high and was in favour of neck abscess. FNAC from a diffuse swelling yielded approx. 07 ml thick pus with dense mixed inflammatory cells infiltrate comprising of predominantly neutrophils with lymphocytes , plasma cells and eosinophills on a background containg cell debris, necrotic material with scattered cystic macrophages which ruled out any thyroiditis as there was no parent thyroid cells. Contrast enhanced CT scan (Fig No.2) showed a large thin peripherally enhancing lesion with central cystic/ necrotic area & a few air pockets in neck antero- inferiorly involving anterior strap muscles ,traversing the left lobe of thyroid , indenting the left sternocleodomastoid muscle with mild overlying subcutaneous edema. Posteriorly, the lesion extended into the retropharyngeal space, posterior hypopharyngeal wall towards the left and laterally into the left carotid space. The retropharyngeal edema extends superiorly upto C1. Reversal of cervical curvature noted with no bony erosion. Mild edema was seen in walls of left pyriform sinus. Mildly enlarged lymph nodes were seen at levels I, II, & V bilaterally. CT Scan with oral contrast revealed a tract starting from left Pyriform Sinus and crossing the left thyroid lobe. Patient was first managed conservatively with broad spectrum antibiotics to which he responded very well. No incision and drainage was required and patient was discharged in satisfactory condition. He was later called for follow up after a course of adequate oral antibiotics and anti-inflammatory drugs. Flexible laryngoscopy was done which revealed a sinus opening originating from the left side of the pyriform sinus apex (Figure No. 3).Patient was taken up for direct laryngoscopy and exploration of sinus under general anesthesia. Hypopharyngoscope was inserted in left pyriform fossa, opening of sinus visualized and chemical cauterization was done. Patient was put on ryles tube feeding for five days and discharged in satisfactory condition. Patient is in regular follow up and for the last two months he is asymptomatic.

DISCUSSION

Acute suppurative thyroiditis and thyroid abscess are extremely rare disorders. A rich blood supply, a generous lymphatic drainage, a high iodine level that inhibits bacterial growth, and a complete, protective fibrous capsule are factors that have been mentioned as contributing to the relatively high resistance of the thyroid gland to infection[6].Congenital pyriform sinus fistula has recently been recognized as an underlying cause of acute suppurative thyroiditis or acute deep neck infection[7]. The combination of a congenital sinus and acute suppurative thyroiditis was first described in the Japanese literature[8]. In 1973, Tucker and Skolnick described a patient with a recurrent left-sided neck abscess in whom a fistulous tract from the apex of the pyriform sinus to the lateral part of the neck was found[9]. Several other, similar cases have been reported subsequently, which are now believed to be infections arising from an underlying congenital pyriform sinus fistula, probably a remnant of the third or fourth branchial pouch[10,11]. The diagnosis of a persistent pyriform sinus requires an index of suspicion. A recurrent left neck abscess or acute thyroiditis that does not respond to appropriate antibiotic and surgical therapy should raise suspicion of this entity. Fine-needle aspiration biopsy should be the initial diagnostic test, especially when malignancy is being considered. Culture of the aspirate should be done to guide antimicrobial therapy. Barium swallow has been advocated as an effective means of demonstrating the presence of an anomalous tract. However, false negatives can occur due to soft tissue oedema in the acute phase. The oedema causes closure of the tract. Edema occurs during acute infection and may prevent contrast medium from entering the sinus tract [12]. Thus, the optimal time for this study is the quiescent

stage after antibiotic therapy. Further confirmation of a sinus tract may be obtained by direct laryngoscopy[13]. Computed tomography scan is the technique of choice to assess the extent of infection [14].

CONCLUSION

The differential diagnosis of a child with acute neck swelling is varied and the presence of an underlying pyriform sinus fistula is often overlooked, resulting in recurrent infections. A history of preceding upper respiratory tract infection and left- sided neck infection should alert the physician to its presence. Ultrasound and CT demonstrate neck infections well and can depict the presence of a fistula. Barium oesophagography helps to confirm the presence of the pyriform sinus fistula and delineates its extent prior to surgical treatment. Combining a proper preoperative evaluation with careful surgical planning may result in the proficient eradication of these lesions, offering the patient relief from this source of recurrent infection. Definitive management is achieved by complete surgical excision of the anomaly.

Fig.No.1 Showing left sided swelling involving thyroid region.

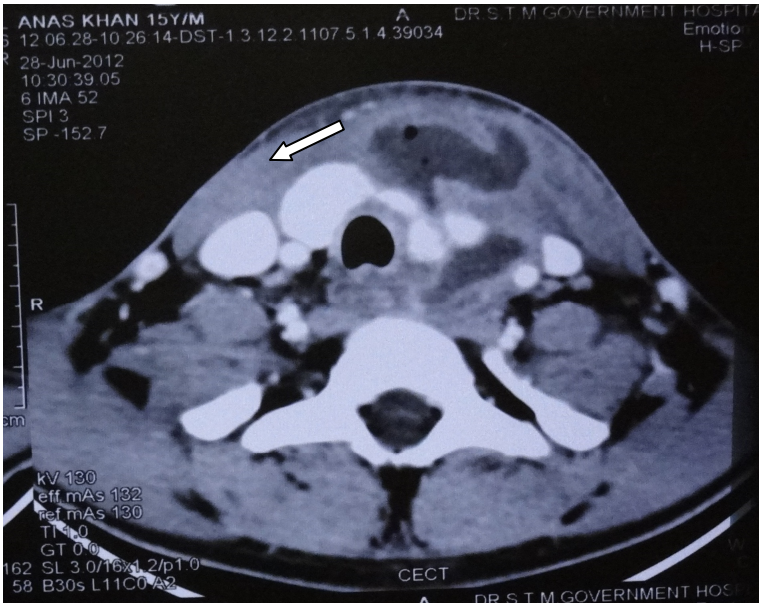

Fig No.2 CECT Scan neck showing extent of abscess with arrow pointing to air present in the lesion.

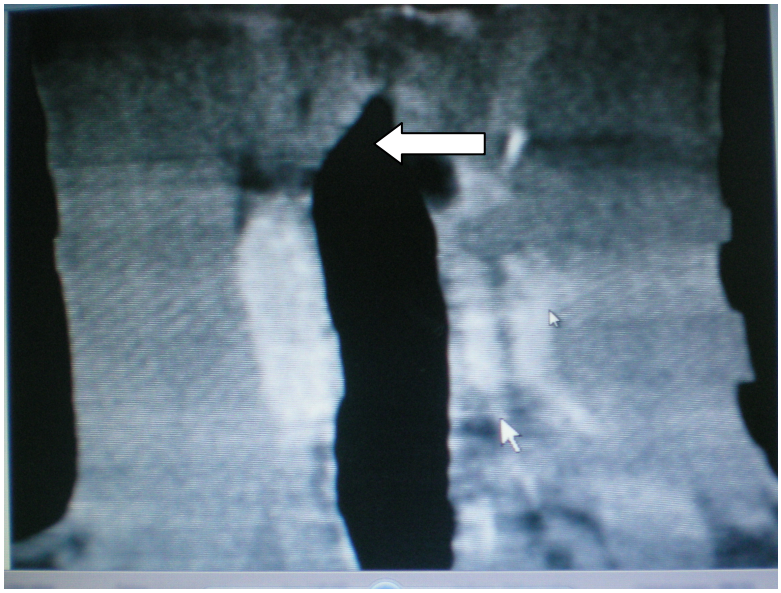

Fig No.3 CECT Scan neck showing linear tract (small arrow),Upper large arrow showing contrast in the tract.

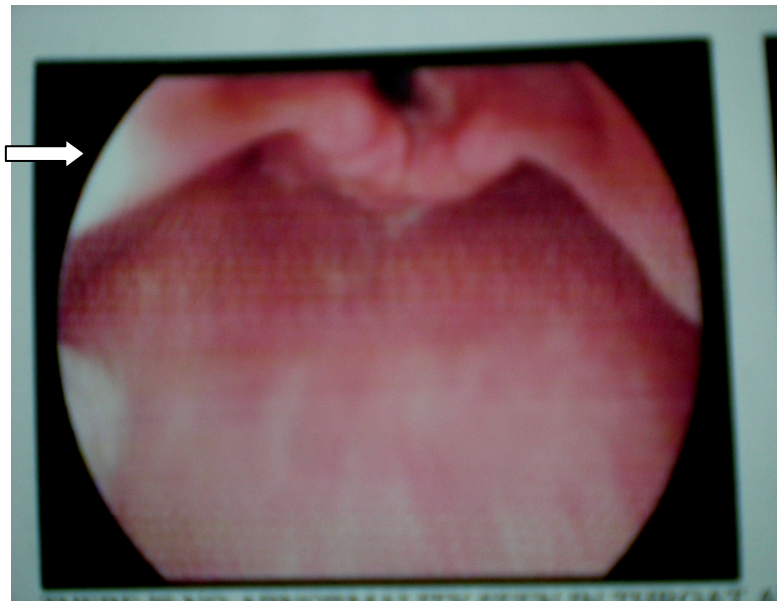

Fig No.4 Solid Arrow showing an opening in left pyriform fossa

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