Double Thyroglossal Duct Cyst Derived from a Single Tract: A Case Series

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ABSTRACT

Thyroglossal cyst is one of the common midline cystic lesions of neck attending paediatric surgery clinic. However double cyst is anatomically rare and sparsely described clinically in all searched English literature. We have retrospectively collected and described three cases of double cyst from our clinical practice. Two of three cases were females of age below ten years and one boy of thirteen years of age who presented with complaint of swelling in midline of anterior aspect of neck. There were neither pain/discharge from the swelling nor difficulty in deglutition in all cases. Parents had noticed swelling in infancy or early childhood which had attained to a size so as to cause cosmetic issues in girls. In the contrary, the teen male was having redness over the swelling one year back which subsided on symptomatic treatment but continued to increase in size. Ultrasonography was done in all cases to delineate normal thyroid tissue, and two of three cases had a significant second cystic lesion suprahyoid, proximity to hyoid bone. Planned for excision of lesion under general anaesthesia. In all cases, we found two cystic swelling at both ends of single duct tissue. After careful dissection and excision, closure of wound was done in a cosmetic manner and tissue on histopathology confirmed to be thyroglossal cyst at both ends. On follow-up, all cases were doing well without recurrence. Surgeons should be vigilant not to ignore any cystic swelling if found during Sistrunk operation and it should be carefully excised.

Keywords: Thyroglossal cyst, Thyroglossal tract, Sistrunk

INTRODUCTION

Thyroglossal cyst is one of the most common surgical problems of the neck encountered by pediatric surgeons. Thyroglossal cyst with other cysts of second branchial arch most common the cystic lesions of the neck. [1-2] The thyroglossal cyst develops due to collection in the remnant of the thyroglossal duct. The thyroglossal tract descends from the base of tongue, foramen caecum and end below the hyoid bone developing onto the two lobes of thyroid. Due to typical close relation with the hyoid bone the excision is associated with recurrence if the part of bone is not excised. But developmental anomalies like double cyst form single duct have also been reported. [3-4] These cases are extremely rare and can lead to recurrence if both lesions are not excised. We present our experience of three cases with duplicated cyst arising from single thyroglossal tract.

Patient details and method

Case 1: Eight year girl presented to OPD with complaint of swelling of neck for last six years without any pain, tenderness or discharge from swelling. Parents noted progression of swelling from the time they first noticed it. There was no associated systemic complaint. Swelling was tense cystic & mobile on protrusion of tongue.
Ultrasonography was advised to rule out ectopic thyroid tissue in the cystic swelling and to ascertain anatomy of bilateral thyroid lobes. With all routine investigation and serum thyroid profile, Sistrunk operation was planned and on table post excision of hyoid, dissection was carried through the mylohyoid, the tract was found to be in continuity with another cyst in suprathyroid region which was comparatively smaller than infrahyoid cyst. [Figure 1] There was evidence of a connection with the base of tongue/foramen caecum by duct strand. Wound was closed in layers with skin by subcutaneous absorbable suture. Tissue was sent for histopathology confirming cyst to be lined by stratified ciliated squamous epithelium with occasional giant cells. On follow-up, our girl patient is doing well without recurrence and healthy wound line.

Case 2
A school going male child of thirteen years of age referred to us with complaint of swelling of neck which was prominent for last one year. There was history of redness with tenderness which subsided with local practitioner treatment. Progressive increase in size concerned parents assuming malignancy. They visited otorhinolaryngology outpatient for the same and referred to us. Ultrasonography of neck and routine investigation were performed, planned for Sistrunk operation. Sonography commented about small cystic swelling near floor of mouth in close proximity to hyoid in addition to infrahyoid cystic swelling with thick consistency content along with base of tongue free. Intraoperatively, cyst was dissected and haemostasis was maintained using bipolar cautery.[Figure 2] The hyoid space was occupied by small soft fluctuant swelling which was extending to floor of mouth. Meticulous dissection was continued to delineate cystic margin and excision of intact cyst was done. Rest of surgery and postoperative period was uneventful with last follow-up done at six months having good results.

Case 3
We encountered a case in routine paediatric surgery outdoor of a girl of age seven years with chief complaint of small swelling in midline neck since one year of age. It was not increasing in size and on palpation felt bigger than clinically visible. On protrusion and deglutition, it was mobile confirming it to be thyroid remnant.[Figure 3] No other significant history was noted. Thyroid serum profile was in normal range. Ultrasonography commented about a 1.5x2.5centimeter cystic lesion with normal bilateral lobes of thyroid which was situated in infrahyoid region. It added also a cystic to firm mass of 0.5x1.0centimeter lesion just posterior to body of hyoid with high probability of a lymph node of cervical group. After routine pre-operative workup, patient was posted for surgery and on
dissection, we found a cystic lesion in infrahyoid part and in continuity of tract one more cystic lesion dorsal to hyoid bone. Excision of cystic mass along with segment of body of hyoid bone was done with closure of wound achieving possible cosmesis. Histopathology confirmed both cyst being lined by respiratory epithelium and thyroglossal cyst.

**DISCUSSION**

Thyroid gland develops from thyroglossal tract arising from the foramen caecum and so has pharyngeal connection. The tract usually involutes by 9-10th week of gestation. The failure of the involution and secretion form the epithelial tract leads to the formation of the thyroglossal cyst.\(^4\-5\)

Due to the embryological origin, the tract descends very close to the thyroid cartilage in close relation to the hyoid bone and in most cases lie in the sub hyoid location. The cyst can be in very close relation to the underlying thyroid gland. Some cyst can have only thyroid tissue in the neck i.e. ectopic thyroid. Sonographic preoperative screening of normal bilateral lobes is essential before excision of the cyst. In cases with cyst in close relation to the thyroid lobe, deeper exploration with complete excision of the cyst is required to prevent recurrence. Mc Henry et al reported a cyst that adhered to the left thyroid lobe. The cyst stretched as far as the hyoid cartilage and was managed by excision of cyst followed by Sistrunk procedure.\(^5\)

In other reported cases the cyst extended in different regions like suprathyroid region, intrathyroidal region and sometimes intra-hyoid region. But none of the series or cases form except one series reported duplicated cyst form one tract. Our series was unique as its largest series of this kind giving insight to the management of such cases avoiding complication of the recurrence.\(^6\)

A careful clinical history and physical examination is extremely important in diagnosis. On examination one can feel two cystic lesions moving with each other on palpation. Ultrasound is extremely useful for the diagnosis and to visualize the normal lobes of thyroid.

In atypical lesions the fine needle aspiration cytology can help to differentiate the lesions from cold thyroid nodule.\(^7\) Complete excision with Sistrunk’s operation is treatment of choice.\(^8\)

**CONCLUSION**

The duplicated cyst from single tract is extremely rare and careful history, diagnosis and complete excision of both the lesion is important to avoid recurrence.

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**REFERENCES**
