Presternal Thyroglossal Fistula: A Rare Case Report

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ABSTRACT

Thyroglossal cyst is a congenital condition of the neck where the painless swelling is found in the midline of the neck in between the foramen cecum of tongue base and sternal notch. Condition is common in children.

Thyroglossal fistula is either secondary to infection or drainage of a misdiagnosed abscess.

Here we are presenting a case of thyroglossal fistula with its opening over the chest and a cord extending from the hyoid bone to the chest causing restriction of the neck movement. Thyroglossal fistula opening in the chest, i.e. over the sternum is very rare and not been reported in any literatures.

Treatment of this type of thyroglossal fistula is same as other types, i.e. Sistrunk's operation, where tract along with part of the hyoid bone is to be removed to prevent recurrence. We are reporting this case for its rare occurrence.

Keywords: Thyroglossal fistula, Foramen cecum, Hyoid bone.

How to cite this article: Rout MR, Mohanty D, Bobba K, Meta C, Karri S. Presternal Thyroglossal Fistula: A Rare Case Report. Int J Head Neck Surg 2013;4(2):92-94.

Source of support: Nil

Conflict of interest: None declared

INTRODUCTION

Thyroglossal duct cysts are the most common congenital cysts in the neck. Most patients present are children, although presentation at any age is possible. Male and females are equally affected, and the cysts are usually asymptomatic but they may become infected and form abscesses and draining fistulas.

Thyroglossal duct cysts are cystic dilatations of epithelial remnants of the thyroglossal duct tract, just formed during the migration of the thyroid during embriogenesis. During its migration the gland remains connected to the tongue by a narrow canal, the thyroglossal duct.

Thyroglossal duct cysts can arise anywhere there has been failure of the complete obliteration of the tract, the commonest site being just above or just below the hyoid bone. Cystic dilations of this tract remnant result in the clinical presentation of a midline neck mass.

A thyroglossal fistula is usually caused by an attempted drainage of a misdiagnosed abscess or to an inadequate excision leaving the hyoid bone intact.¹

So, usually thyroglossal fistula is found in between the area of thyroid gland (sternal notch) and foramen cecum of tongue base.

CASE REPORT

A 10 years old child came to our OPD with chief complaints of a discharging sinus over the chest since 2 years and restriction of neck movement since 2 years. It started with a small cystic lesion just below the thyroid cartilage 6 years back with slowly increasing in size. Gradually it descended down below the sternal notch and presented over the body of the sternum. Two years back it has been drained by some local practitioner and since then the cyst became a fistula and discharging intermittently. There was restriction of neck movement.

On examination there is a sinus in the chest over the body of the sternum 2 cm below the suprasternal notch with granulation tissue. On palpation there was a thick cord like structure extended from the sinus up to the level of hyoid bone. Movement of the cord and sinus opening was appreciated during the swallowing and protrusion of the tongue. There were no other swellings or lymph nodes in the head and neck region.

All routine investigations were within normal limit including chest X-ray. Mantoux test was negative. Thyroid function test (T3, T4 and TSH) was normal. Ultrasonography of the neck showed the tract extending from the hyoid bone to sternal body without any communication to thorax or any deeper structures. Thyroid gland was there in normal anatomical position. Fistulogram showed a very small tract just over the sternal body. FNAC of lesion showed only chronic nonspecific inflammation without any evidence of malignancy or tuberculosis.

A provisional diagnosis of thyroglossal fistula was done and planned for Sistrunk's operation.

For the excision of tract three horizontal incisions were given, one at the level of hyoid bone, second at the level of thyroid gland and third oval incision in area of fistula over the sternal body. Dissection started from the sternal body toward the hyoid bone. At last the hyoid bone was exposed and body of the hyoid along with a triangular piece of soft tissue was excised.

After excision the tract and part of hyoid bone measuring approximately 8 cm was sent for histopathological study (Figs 1 to 3)

Histologically, the lesion was described as a cyst wall lined by squamus epithelium and pseudostratified columnar epithelium with hyalinization. Multinucleated giant cells were also present. Thyroid tissue was identified in the specimen. The histopathologic diagnosis was consistent with thyroglossal fistula.

Postoperatively patient's neck movement was improved. There was no evidence of recurrence till 1 year.

DISCUSSION

Thyroglossal duct/fistula presents the most common congenital anomaly in the neck.² It accounts for 70% of congenital neck abnormalities³ and 2 to 4% of all the neck masses.⁴ It results from retention of the epithelial tract between the thyroid gland and its origin, the foramen cecum. Formation of the cyst is likely due to continuous mucus production from the glands found in the duct.

The lesion usually presents as a painless swelling in the midline or paramidline of the neck. The classic description of the lesion is that of a painless swelling in a young child along the midline of the neck which rises with deglutition or tongue protrusion. The lesion is compressible and may fluctuate in size. If, however, it is entwined with the hyoid



Fig. 1: Tract of thyroglossal fistula

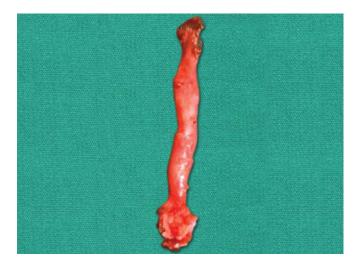


Fig. 2: Thyroglossal tract along with body of the hyoid bone

bone, it may not display any movement with tongue protrusion or swallowing.⁴

Thyroglossal cysts occur in six different variants; infrahyoid, suprahyoid, juxtahyoid, intralingual, suprasternal and intralaryngeal cysts (extremely rare). Most commonly it is present in first decade of life. However, they are seen in adult also. 6

Delineating the different types of cysts is important since the management of thyroglossal duct cyst requires not just simple excision but rather a Sistrunk operation which requires excision of the cyst, the central portion of the hyoid bone, and removal of a core of muscles up to the base of the tongue. Failure to do so may lead to recurrence of the cyst.

The histologic appearance of a thyroglossal duct cyst is a cyst lined by respiratory epithelium with thyroid tissue, mucus glands, and small patches of lymphoid tissue variably present in the connective tissue wall. The presence of thyroid tissue in the connective tissue wall of the cyst is considered pathognomonic of thyroglossal duct cyst, however not all specimens display such tissue.

The presence of a nodular density in the thyroglossal duct cysts suggests the possibility of an associate carcinoma. The incidence of carcinoma associated with thyroglossal duct cysts is low (less than 1% in literature).⁷

CONCLUSION

Thyroglossal duct cysts are the most common congenital cysts in the neck. Thyroglossal duct cysts can arise anywhere there has been failure of the complete obliteration of the tract, the commonest site being just above or just below the hyoid bone. Cystic dilations of this tract remnant result in the clinical presentation of a midline neck masses. A thyroglossal fistula is usually caused by an attempted drainage of a misdiagnosed abscess or to an inadequate excision leaving the hyoid bone



Fig. 3: Postoperative picture

intact. Thyroglossal fistula over the sternum is rare and not been reported in any literature. This case has been reported because of its rare occurrence.

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