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Case Report

Recurrent infected thyroglossal sinus: a unique case

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ABSTRACT

Thyroglossal sinus are a frequent clinical entity in the pediatric population, mostly present in upper midline of the neck. It is the most common complication following thyroglossal duct cysts and is well known for recurrence, if inadequately excised or infected. A careful consideration of the magnetic resonance imaging (MRI) pre-operatively helps improve the outcomes and reduces complications. We present the case of a 12-year-old boy operated four times for recurrent thyroglossal sinus. The diagnosis was confirmed by pre-operative history, physical examination as well as post-operative histopathological report of the specimen. MRI neck was used for tract diagnosis.

Keywords: Sinus, Thyroglossal, Recurrence, Imagine

INTRODUCTION

The thyroglossal duct cysts (TGDC) are the most common congenital neck mass present in the pediatric population. It develops due to the partial involution of thyroglossal duct which is formed in the path of descent of the thyroid gland, from the foramen caecum to its normal anatomical position in the lower part of neck. Hence, the cyst may develop anywhere from the foramen up to the thyroid isthmus or pyramidal lobe. Since these cysts are lined with the squamous or pseudostratified columnar epithelium of secretory nature with or without presence of ectopic thyroid tissue, they are more vulnerable to infection and inflammation.[1] The infection of the cyst or its incomplete excision may cause it to make an external communication with neck skin. Once it develops an internal communication with foramen cecum, it is known as thyroglossal fistula (TGDF). Otherwise, designated as thyroglossal sinus (TGDS).[2] Both TGDS or TGDF generates management difficulties due to inoculation of epithelial cells in surrounding tissue. Hence, incomplete excision with standard surgical procedure is expected. Recurrences are more common in TGDF and TGDS than the TGDC. The Sistrunk procedure is considered as the gold standard for this disease. It is defined as the removal of sinus tract up to foramen caecum and the body of hyoid bone. The removal of hyoid body is an important step of as the tract may pass under, within or over the hyoid bone, this reduces the recurrence rate from 80 percent to 3 to 6 percent.[3] These recurrences may be due to deviation of tract tissue more laterally or may be implantation of thyroglossal epithelial cell in nearby surrounding tissue during phase of infection or surgery.

Ultrasound, computed tomography (CT) and magnetic resonance imaging (MRI) play a supplementary role in diagnosis of these cases, assessment of anatomical extent and complications as well for planning the treatment. MRI is especially useful to identify branched and polycystic thyroglossal duct cysts and demonstrate fluid content. It is the preferred modality to identify lesions close to tongue base.[1,3]

Here, we are presenting a case of TGS which was refractory to surgical excision, including Sistrunk procedure. We did a dynamic MRI to evaluate the residual tract and tissue before exploration. It was successfully excised, being posteriorly placed in base of tongue rather than foramen cecum. Hence, our case is unique to emphasize the role of dynamic MRI as a choice of preoperative imaging for the planning to manage a TGDC, TGDF or TGDS.

CASE REPORT

A 12-year-old boy presented to ENT outpatient department of our hospital with complaints of intermittent discharge from an external opening in the upper part of the neck for last 5 years. Further history revealed that 5 years back, the child had developed an abscess at a point just lateral to midline of neck at the junction of upper one third and lower two third which ruptured spontaneously after 10 days. Intermittent discharge followed the rupture of the abscess, following which the child was operated twice at peripheral institutions. On first contact at our centre, the patient presented with an external opening in the upper one third of neck had intermittent discharge which was scanty, mucopurulent, not blood tinged, not associated with pain. The gold standard, sistrunk procedure was performed on the patient but there was reappearance of the sinus on the tenth day postoperatively, a MRI neck to delineate the sinus tract was performed, the findings were hyperintense collection of size 20×18×23 mm noted in sublingual region collection was seen reaching up to the base of tongue. A well-defined sinus tract was delineated at the posterior aspect of base of tongue (Figure 1).

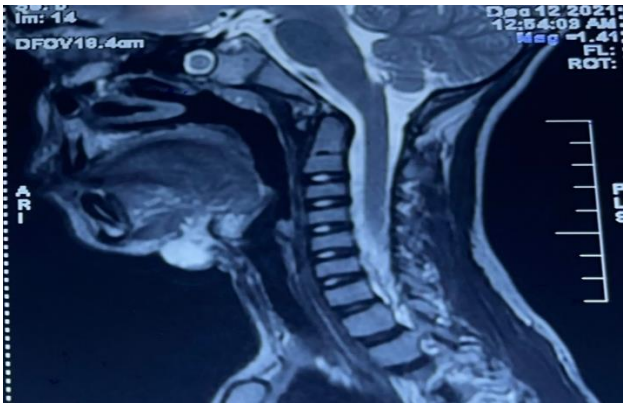

Figure 1: Sagittal cut of the MRI shows thyroglossal remnant posterior in base of tongue. The sinus tract is also revealed.

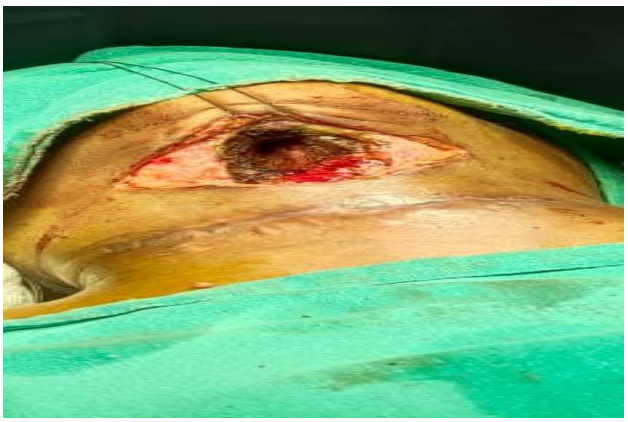

Figure 2: Intraoperative picture shows wide anterior local excision being performed of recurrent thyroglossal sinus.

Subsequently, anterior local wide excision was performed under general anesthesia with proper consent (Figure 2). Thyroid status of the patient was evaluated by TSH, T3 and T4 levels. Postoperatively, the wound healed well and the histopathological report confirmed infected thyroglossal duct tissue. On follow up, patient reported complete recovery.

DISCUSSION

Thyroglossal duct cyst recurrences are attributed to incomplete excision of the thyroglossal tract and its branching ductulus in 8% cases. Inflammation or infection in the form of an abscess is the most common complication in TGDC patients. Risk factors for recurrence include rupture of cyst, post-operative infection, and limited experience of the surgeon and persistence of infra or suprahyoid tract remnant.[4] As our case shows, TGDF and TGDS often present with management difficulties due to inoculation of epithelial cells in surrounding tissue. Also, proper radiological evaluation with aids like MRI if not done preoperatively, can be associated with recurrence as well. Often, hyoid bone is left or not excised which also increases the recurrences.[5]

Definitive treatment remains the Sistrunk operation. Recurrence rate is 6.6% in pediatric population irrespective of the surgery performed, Sistrunk or modified Sistrunk operation.[3,5] Central neck dissection or anterior wide local excision is also performed to remove the entire thyroglossal tract remnant ,this reduces the risk of initial failure and is considered a favourable option in management of recurrent cases.[5] In this patient, since the tract was confined to the posterior part of tongue with extension of cystic lesion in anterior cutaneous plane, we made sure to completely excise the tract up to base of tongue, along with the removal of the hyoid bone in its entirety ,to avoid any chance of residual tract. Central neck dissection had no role in the management as the remnant was posterior to the base of tongue.

Appropriate antibiotic coverage was given with placement of a drain. Stitches were removed on the seventh day, the wound completely healed and patient achieved complete cure. This case is novel in terms of using the advanced radiological aids like MRI, a departure from the classical fistulograms to identify the residual disease and successfully using it to achieve cure.

CONCLUSION

The thyroglossal cyst, sinus or fistula is notorious mid-line congenital neck pathology, if not managed properly, is known for recurrence. The dynamic MRI has especially beneficial to locate the lesion within the normal soft tissue of the neck. This amount of knowledge through MRI is highly beneficial to determine the extent of disease tissue excision to prevent recurrences.

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