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RESEARCH ARTICLE

THYROGLOSSAL DUCTAL ANOMALIES IN CHILDHOOD: 16 YEARS EXPERIENCE AT A TERTIARY CARE CENTRE

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ABSTRACT

Background: Thyroglossal ductal anomalies classically manifest as painless midline anterior neck swelling. Diagnosis is usually based on clinical presentation.
Objective: The aim of present study is to review cases diagnosed with thyroglossal ductal anomalies and the subsequent approach to management.
Methods: The medical records of patients diagnosed with thyroglossal duct cysts and fistula from January 2001 till June 2016 were retrospectively analysed.
Results: A total of 43 patients were included of which 28 were males and 15 were females accounting for male to female ratio of 1.9:1. The age ranged from 4 days to 15 years. Twenty three cases (53.48%) presented with discharging thyroglossal fistula while twenty cases(46.51%) as a painless cystic neck swelling. The most common site of the cyst was infrahyoid in 25 cases (58.14%).
Conclusion: Thyroglossal ductal anomalies should be diagnosed and managed surgicall yearly to prevent fistula formationand recurrence. Sistrunk operation is the surgery of choice at our centre.

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INTRODUCTION

Thyroglossal ductal anomalies are the most common congenital/developmental neck swellings in childhood, representing more than 75% of midline neck masses. Thyroglossal duct cysts often present during childhood by the age of 10yrs, and these are the second most common benign neck mass after lymphadenopathy in paediatric age group (Thabet, 2011). However, these can also present later in adulthood. Thyroglossal duct cystsoriginate from persistent epithelial remnants of the thyroglossal tract. Thyroglossal tract usually obliterates by the tenth week of gestation. During the descent of the thyroid gland from the foramen cecum to its final position in the anterior neck, persistent portions of the tract and associated remnants of thyroid tissue presents characteristically as a midline neck mass often closely associated with the hyoid bone (Mondin, 2008). The most common location for a thyroglossal cyst is midline neck close to hyoid or slightly off midline. Approximately 10% - 24% of the cysts are located slight laterally usually on the left (Pollock *et al*., 1966). The cyst is most commonly seen at infrahyoid position in about 25-65% of cases, in suprahyoid location in 20-25% and at the level of hyoid bone in 15-50% cases (Mittal, 2012).

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The cysts located in the neck have lining similar to thyroidal acinar epithelium while those situated near the tongue are lined by stratified squamous epithelium. There is an increased incidence of ectopic thyroid tissue seen in association wththyroglossalductal anomalies (40%). For removal of cyst, Schlange in 1893 first proposed excision of the cyst. Recurrence rate was 20% with this procedure and later in 1920, Sistrunk advised removal of cyst along with core of hyoid bone, reducing the recurrence rate to 3%.[13] In Sistrunk's first series which was reported by Bennett *et al*, recurrence rate was also 3%. Thyroglossal duct cysts can get infected and can be misdiagnosedas abscess leading to inadequate surgical excision or it may rupture spontaneously resulting in fistulous tract known as thyroglossal fistula.

MATERIALS AND METHODS

This study presents a retrospective analysis of patients diagnosed with thyroglossal ductal anomalies during the period from January 2001 to June 2016 at PGIMS, Rohtak. The available records were reviewed for demographic analysis, clinical and radiological assessment, and surgical interventions and histopathology.

RESULTS

Forty three patients with diagnosis of thyroglossal duct anomaly were included in our case series. Out of these, 23

cases were diagnosed with thyroglossal fistulas (53.48%) and 20 cases with thyroglossal cysts (46.51%). Their age varied from 4 days to 15 years with majority of them in age group between 8-10 years (Table 1). Mean age of presentation was 7.15 years. There was a male preponderance with 28 males (65.11%) as compared to 15 females (34.88%) with sex ratio of 1.9:1. Most common position of cyst was infrahyoid in 25 cases (58.14%) followed by suprahyoid in 13 cases (30.23%), juxtahyoid in 4 cases (9.30%) and suprasternal in 1 case (2.32%).

Table 1. Age distribution of cases

Age group (in years)	Number of cases
0-2	3
2-4	6
4-6	7
6-8	6
8-10	8
10-12	7
12-14	5
14-16	1

Clinically, all cases showed the classical presentation of a cyst in the anterior neck with mass moving with deglutition and protrusion of the tongue. The range of duration of the symptoms varied from one month to 10 years with the mean duration of the symptoms is 12 months. The size of the cysts varied from 1.4 to 4.0 cm in diameter. Investigations included radiological evaluation i.e. ultrasonography, FNAC and thyroid profile. USG revealed the classical presentation of well defined, smooth, hypoechoic lesion with posterior acoustic enhancement separate from the thyroid gland with normal appearance of the thyroid gland in all cases of thyroglossal duct cyst. However, cystic lesions were not seen in cases with fistula. Fine needle aspiration documented benign squamous cells and mucoid proteinacious material adding in confirmation of thyroglossal duct cyst.The standard surgical procedure Sistrunk operation was performed in all cases of cyst which involved the radical excision of the wholetract of the duct and cyst along with the body of hyoid bone. In cases with cutaneous fistula an island of skin was also removed together with the duct and cyst. Post-operative histopathological evaluation of the excised specimens confirmed the diagnosis ofthyroglossal duct cyst. Recurrence was seen in 2 cases as core of hyoid bone was not removed in these cases and wound infection was observed in 2 cases which was managed during follow up.

DISCUSSION

Various differential diagnosis can be made for congenital anterior midline swelling but the most common is thyroglossal ductal anomalies in the neck (Hsieh, 2003). Dermoid cyst, branchial cleft cyst, ectopic thyroid, lipoma, sebaceous cyst, metastatic thyroid carcinoma and enlarged lymph node are the important differential diagnosis of thyroglossal ductal anomalies that need to be kept in mind (Moorthy, 2011). It has been reviewed in literature that thyroglossal duct may have a bimodal age distribution with peaks at 6 and 45 years of age (Brousseau, 2003). In our study age ranged from4 days to 15 years with mean age of 7.15 years. Previous studies report thyroglossal duct cyst is more common in males (Thabet, 2011). In our study also, majority of patients were males accounting for 65.11% (28 cases). Thyroglossal ductal anomaly typically presents as mobile midline anterior neck swellingin proximity to the hyoid bone which moves with deglutition and protrusion of the tongue. The most common location reported in literature is infrahyoid. Allard reported 381 cases of diagnosed thyroglossalduct cyst and as regards the site 60.9% were infrahyoid, 24.1% suprahyoid, 12.9% suprasternal and2.1% lingual (Allard, 1982). Thabet *et al*. reported 91% cysts to beinfrahyoid in position, suprahyoid and lingual thyroglossal cysts 4.5% each and nojuxtahyoid or suprasternal cysts were reported in their study on thyroglossal duct cysts (Brousseau, 2003). According to our analysis most common site involved was also infrahyoid (58.14%) followed by suprahyoid (30.23%), juxtahyoid (9.30%) and suprasternal (2.32%). In one of our case, ectopic thyroid was misdiagnosed as thyroglossal duct cyst clinically. However, FNAC was done for confirmation before excision which revealed thyroglossal duct cyst. In a study of 1534 cases by Allard, 32.6% presented with fistula (Allard, 1982). In the other case series by Ostlie *et al*, 33%of cases presented with abscess leading to fistula formation (Ostlie, 2004). In contrast, our analysis revealed 53.48% cases with thyroglossal fistulas and 46.51% with thyroglossal cysts. In most instances, diagnosis depends on clinical presentation. However, USG remains the initial imaging diagnostic modality of choice, especially in children as it is easily available, inexpensive, does not involve sedation or ionizing radiation, and also provides all necessary information for diagnosis and management (Ahuja, 2000). Sonographic demonstration of a normal thyroid gland is necessary to exclude ectopic thyroid in patients with thyroglossalduct anomalies (Moorthy, 2011). MRI T2-weighted image is the only sure imaging investigation as it also visualizes the complete tract to the tongue base. Therefore, MRI is crucial in the diagnostic workup of thyroglossal ductal anomalies with atypical presentation (Thabet, 2011). Dermoid cyst, branchial cleft cyst, ectopic thyroid, lipoma, sebaceous cyst, metastatic thyroid carcinoma and enlarged lymph node are the important differential diagnosis of thyroglossal ductal anomalies that need to be kept in mind (Moorthy, 2011). In our study standard surgical approach, Sistrunk operation was performed in all cases with thyroglossal ductal cyst which involved the resection of body of the hyoid bone together with the whole length of the duct and cyst. An island of surrounding skin was also removed along with the track in cases with thyroglossal fistula.

Conclusion

- Thyroglossal duct anomalies are the most common congenital midline neck swelling that may present at any age but incidence is higher in paediatric age group.
- Thyroglossal ductal cyst can be misdiagnosed as abscess leading to spontaneous rupture or iatrogenic fistula formation. Differential diagnosis of any anterior neck swelling must include thyroglossal duct cyst.
- Thus, any midline neck swelling should be diagnosed and managed as early possible.

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