Thyroglossal Duct Cyst—More Than Just an Embryological Remnant

Sujatha Narayana Moorthy · Rekha Arcot

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Abstract Thyroglossal duct cyst is a congenital malformation that occurs due to incomplete closure of the thyroglossal duct. Apart from being a quiescent embryological remnant, it presents itself clinically at any age and often requires surgical excision. Twenty four patients were encountered at Sri Ramachandra Medical College and Research Institute between June 2004 and June 2009 with thyroglossal duct cyst. The purpose of the study was to analyse their presentation, associated complications including malignancy and the approach to their management. Operative notes, histopathology files and medical records were used for the retrospective analysis of the patients with thyroglossal duct cyst. Thyroid profile and Radioisotope scan were performed on all the patients to document the position of the thyroid gland. Further, these patients were subjected to Ultrasonography and Computerised Tomography neck to conclusively map the location of the cyst. Fine Needle Aspiration of the tissue was done to diagnose malignancy. It was noted that there was maximum clustering of about 62.5% in males. About 45% of them belonged to the paediatric age group. The total number of symptomatic patients was about 59%. Unlike the various studies in literature, most of the thyroglossal duct cyst patients at our hospital were symptomatic. The symptoms were pain, dyspnoea, dysphagia, discharge and recurrence. Malignancy was diagnosed in two patients who were ironically asymptomatic. Other complications were looked into and treated. Thyroglossal duct cyst is a cervical anomaly that is usually

found as an asymptomatic painless cystic swelling. However, most of our patients had a symptomatic presentation. Association with malignancy was noted. Surgical intervention was the cure for all the patients.

Keywords Thyroglossal duct cyst · Paediatric males · Symptomatic presentation · Malignancy · Sistrunk procedure

Introduction

Thyroglossal duct cyst (TDC) is the most common type of developmental cyst encountered in the neck region. It is a condition that results from the failure of obliteration of the thyroglossal duct which forms a bridge between the base of the tongue and the thyroid gland. A review of the literature shows that this developmental abnormality arises in about 7% of the population [1]. About 1% of the patients suffering from TDC are noted to have an association with malignancy [1, 2]. Up to now one hundred and fifty cases of TDC carcinoma, predominantly papillary type, have been reported [3]. The ductal cyst often presents as a painless, cystic, mobile, fluctuant and midline swelling of the neck. Interestingly, a unique case was reported by Sauvageau et al. [4], where the TDC caused severe dyspnoea followed by fatal asphyxia leading to patient's death.

The advances in imaging techniques such as Ultrasonography (USG) and Computerised Tomography (CT) of neck are used in the confirmation of TDC and associated carcinoma. Since 1920, the gold standard for treatment has been Sistrunk procedure [5, 6].

Various studies have been done on TDC to understand its varied and wide range of manifestation. Hence the purpose behind this study was to analyse the presentation, associated complications including malignancy and the

S. Narayana Moorthy · R. Arcot
Sri Ramachandra Medical College and Research Institute,
Porur,
Chennai 600116, India

S. Narayana Moorthy (✉)
617, G2, Sri Ramana Apts., 9th Sector, 51st Street, K.K. Nagar,
Chennai 600078, India
e-mail: nsujatha88@yahoo.co.in

approach to management in the patients with TDC in our hospital.

Patients and Methods

The paper presents a retrospective analysis of patients with TDC at Sri Ramachandra Medical College and Research Institute (SRMC & RI) between June 2004 and June 2009. In this five years period, over hundred patients with neck swellings were encountered in the hospital. Out of these, twenty four patients were identified with TDC by perusal of operative notes, histopathology files and medical records.

Thyroid profile and Radioisotope scan were performed on all the patients to check and document the normal positioning of the thyroid gland. Subsequently, USG neck and CT neck were done to map the location of the cyst. Surgical intervention was the cure for all patients. This depended upon the patients' presentation. Sistrunk procedure was performed on most of the patients. In addition to Sistrunk procedure, total thyroidectomy was performed on patients with malignancy. All the patients were followed up for two weeks postoperatively. This period was however uneventful.

Results

Demographics

The demographics in the present study, that reports about 62.5% prevalence of the anomaly in males, indicates that TDC has a predilection for males. Of the patients identified with TDC, about 45% of them were in paediatric age group (Fig. 1). The fact that the most common location of the cyst is infrahyoid, was also brought about in this study, where the TDC was found below the hyoid bone in about 87.5% of the patients (Fig. 2). The various locations of TDC in the patients are shown in Fig. 3.

It was noted that a typical presentation of painless, cystic swelling was found only in about 41.6% of the patients diagnosed of TDC. About 21% of the patients had no symptoms other than pain. Nine patients constituting about 38% had symptoms of hypothyroidism, dyspnoea, dysphagia and watery discharge. Hence the symptomatic patients in this study were about 59%. While it is expected to find majority of the patients to be asymptomatic, the present study non-intuitively suggests otherwise. However, it is to be noted that the number of patients considered in the present study is not sufficiently large to statistically determine the extrapolated characteristics of a general population. About one third of the patients diagnosed with TDC (33.3%) were also found to have complications such as malignancy, infection, inflammation, rupture of cyst, fistula and recurrence.

Fig. 1 The distribution of TDC

Management

Of the twenty four patients, twenty one underwent Sistrunk procedure, of which one patient was found to have a spontaneous rupture of the cyst with watery discharge. Total thyroidectomy was performed on two patients whose histology report suggested a papillary carcinoma (Fig. 4) and its follicular variant. Both the patients were males aged about sixty. Ironically, both the patients were completely asymptomatic despite a huge swelling of 6 cm×8 cm. A thirteen year old patient that was diagnosed of an ectopic thyroid with TDC was managed without surgery. It was noted that this was the only functioning thyroid present.

Discussion

Anatomy

Persistence of non-obliterated thyroglossal duct culminates in the formation of TDC. If the lower part of the duct alone persists, it prevails as the pyramidal lobe of the thyroid. The foramen cecum, which typifies the ductal opening into the tongue, remains a small blind pit in the mid line between the anterior two thirds and the posterior one third of the tongue. This ductal cyst can evolve anywhere along the path of the thyroglossal duct though the possibility of

Fig. 2 The location of TDC

Fig. 3 The various presentations

occurrence within the hyoid bone, within the tongue and floor of the mouth is tenuous.

Ghaneim and Atkin [7] indicated, while TDC is found in between hyoid bone and the thyroid cartilage in about 60% of the patients, it is suprahyoid, supra-sternal and intra-lingual in about 24%, 13% and 2% respectively. However extremely rare locations have also been reported. For instance, Tas et al. had reported an intrahyoid location of TDC [8] and Soon et al. had reported a case of TDC that descended into the mediastinum [9]. Histologically, it is known that the cyst is lined by stratified squamous or pseudo stratified ciliated columnar epithelium.

Clinical Features and Management

Unlike the studies presented in the literature that have shown that majority of the patients come with a painless, smooth and cystic swelling, the patients at SRMC & RI was mostly symptomatic. The symptoms present were pain due to infection, and dyspnoea, dysphagia, dyspepsia which is attributed to the compression of the underlying structures by an unusually large cyst. Watery discharge from a fistula was also noted. A TDC will exhibit movement with protrusion of tongue and deglutition. The differential

diagnosis of TDC includes branchial cleft cyst, lipoma, metastatic thyroid carcinoma, dermoid cyst, sebaceous cyst and enlarged lymph node.

Thyroglossal duct cysts are rarely associated with ectopic thyroid tissue. Occasionally it is also associated with lingual thyroid that presents as a lump at the base of the tongue. It is estimated that only about 1% of the patients with TDC show malignant changes, the most common being papillary thyroid carcinoma. Cases of incidentally diagnosed primary papillary carcinoma of thyroglossal duct cyst have been reported [10]. This is characterised by a non aggressive behaviour and rare lymphatic spread. The other possibilities are follicular variant of papillary carcinoma, squamous cell carcinoma and Hurthle cell carcinoma [11]. Literature has even proved that there can be synchronous occurrence of two malignancies as in both papillary and squamous cell carcinoma in TDC [12]. Inflammation, rupture and recurrence (Fig. 3a) are the other possible complications. The TDC can also rupture spontaneously and manifest as a draining sinus, which is being erringly called a thyroglossal fistula. However, a rare case of complete communication between the neck skin and foramen caecum has been reported [13].

Fig. 4 The thyroglossal cyst with papillary carcinoma and a 10x inset of the papillary carcinoma

In the present study, two cases of malignancy, two cases of rupture with fistula formation, three cases of inflammation and one case of recurrence was noted.

Investigations for the detection of TDC include USG neck, which shows a cystic lesion [14]. When USG was done on all the patients of the present study, two patients revealed the presence of papillary projections, solid components and calcification which raised the suspicion of a co-existing malignancy. Subsequently the diagnosis was substantiated by histopathological examination of the cyst.

CT with contrast shows a well-delineated cystic lesion with capsular enhancement and its relation to the hyoid bone. Radioisotope study is done to document the presence of normal functioning thyroid gland [15].

Earlier, TDC was treated with simple excision or incision and drainage, which resulted in a high recurrence rate of 50% in patients. In 1893, Schlang proposed the excision of the cyst along with the central portion of the hyoid bone, which reduced the recurrence rate to 20% [16].

Treatment of choice now is Sistrunk procedure, in which a portion of the hyoid bone is removed with meticulous excision of the persistent duct up to the foramen caecum. Literature reveals that Sistrunk has brought down the recurrence from 50% to 3% [17]. Shah et al. [18] proved that recurrence of TDC remains low when the Sistrunk procedure is employed. This is independent of presenting age and symptomatology. A ten year old child with recurrence of TDC twice was dealt with in this study. Recurrence was attributed to inaccurate dissection, young age of the patient and intra-operative rupture of the cyst.

Conclusion

Thyroglossal duct cyst is the most common congenital cervical anomaly. While found among all age groups, it was noted that there is maximum clustering in paediatric males, reason being the thinning of adipose tissue during this period of childhood. It was also found that most of our patients were symptomatic thus proving that TDC is indeed more than just an embryological remnant. An association with malignancy was well documented. It is pointed out that knowledge of embryology, meticulous dissection along anatomical planes and removal of the core of the hyoid bone is essential to prevent recurrence of TDC.

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