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MAIN ARTICLE

Wide anterior neck dissection for management of recurrent thyroglossal duct cysts in adults

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Abstract

Objective: Thyroglossal duct cyst recurrence following resection is attributed to anatomical variability and residual thyroglossal ducts. In adults, thyroglossal duct cyst recurrence is extremely rare and a surgical solution is yet to be well explored. This paper describes our approach to the management of recurrent thyroglossal duct cysts and sinuses in adults using a wide anterior neck dissection.

Method: A retrospective review was performed to identify adults who underwent a wide anterior neck dissection for recurrent thyroglossal duct cyst management between 1 January 2009 and 1 January 2015.

Results: Six males and one female were included in the series (mean age, 26.4 ± 10.9 years). Recurrence occurred at a mean of 18 ± 9.8 months following primary surgical management (3 patients underwent cystectomy and 4 had a Sistrunk procedure). All patients subsequently underwent wide anterior neck dissection; there was no further recurrence over the 12-month average follow-up period.

Conclusion: This paper describes a wide anterior neck dissection technique for the management of recurrent thyroglossal duct cysts or sinuses in adults; this approach addresses the variable anatomy of the thyroglossal duct and is associated with minimal morbidity.

Key words: Thyroglossal Cyst; Recurrence; Adult; Neck Dissection

Introduction

Thyroglossal duct cysts are the most common congenital neck mass,[1] and arise in at least 7 per cent of the population.[2,3] Thyroglossal duct cysts typically have a midline locality, with a close relationship to the hyoid bone, and classically move upwards on tongue protrusion.[4] Patients can also present with a draining cutaneous sinus secondary to cyst rupture, which occurs spontaneously or as a result of acute infection.[5,6] Although usually benign, surgical resection of thyroglossal duct cysts and sinuses is recommended in light of the possibility of recurrent infection and the small risk of malignancy.[7]

Thyroglossal duct cysts arise from incomplete involution of the thyroglossal duct, an embryological structure that spans the caudal migration of the developing thyroid gland.[8,9] Throughout this descent, the duct maintains attachments to the gland and its point of origin, the foramen caecum of the tongue base.[8,9] Histological studies suggest that the trailing thyroglossal duct arborises into multiple smaller ductules, all of which usually involute between the 7th and 10th weeks of gestation.[2,10] However, in some individuals, complete obliteration of duct and ductules fails to occur, and persistence of any remnant can give rise to a thyroglossal duct cyst in later life.[11]

Advances in thyroglossal duct cyst and sinus management reflect an increased understanding of the underlying embryological and anatomical bases of development. Following initial approaches of simple cystectomy, or incision and drainage, Schlange described a modified technique that incorporated the excision of the central aspect of the hyoid bone, reducing recurrences to approximately 20 per cent.[12] Sistrunk elected to dissect superiorly and include a core of tissue from the hyoid to the foramen caecum, while ensuring the oral mucosa was not breached.[13] The Sistrunk procedure subsequently reduced recurrences to between 3 and 10 per cent, and is now the accepted first-line method of management.[14–16]

Although wide anterior neck dissection for recurrent thyroglossal duct cysts has been described in the paediatric population,[17,18] occurrence in the adult population is uncommon, and, given the rarity of recurrence, published literature addressing management in this subset

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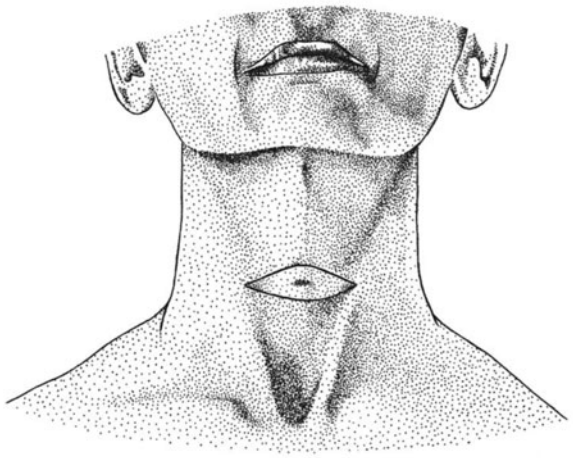

FIG. 1
Elliptical incision surrounding a thyroglossal duct sinus.

of patients is limited. Herein, we describe our experience and surgical technique of wide anterior neck dissection for the management of recurrent thyroglossal duct cysts and sinuses.

Materials and methods

A retrospective chart review was performed to identify adult patients who had undergone a wide anterior neck dissection for the management of a recurrent thyroglossal duct cyst or sinus between 1 January 2009 and 1 January 2015. Electronic medical records and archived paper charts were reviewed to obtain the following data: patient age; gender; date and details of interventions (cyst aspiration, cystectomy, Sistrunk operation); time to, and presentation of, thyroglossal duct cyst or sinus recurrence; details of wide anterior neck dissection; post-operative complications; and outcome at final follow up. Institutional review board approval was obtained (S40046242).

Wide anterior neck dissection

The patient is positioned supine with their neck extended. A transverse incision is performed at the level of the cyst. If a cutaneous sinus is present, a wide elliptical incision is made around the opening of the sinus (Figure 1). Corners of the incision are extended laterally as far as the anterior border of the sternocleidomastoid muscles. Subplatysmal flaps are raised, superiorly to the level of the hyoid bone and inferiorly to the sternal notch.

Dissection begins inferiorly with detachment of the sternohyoid and sternothyroid muscles from their sternal attachment. The dissection proceeds superiorly in a plane beneath the pre-tracheal fascia, and over the trachea, substance of the thyroid gland, cricoid and thyroid cartilages, and thyrohyoid membrane, and continues to the superior attachment of the sternohyoid at the hyoid bone. The thyrohyoid muscle is

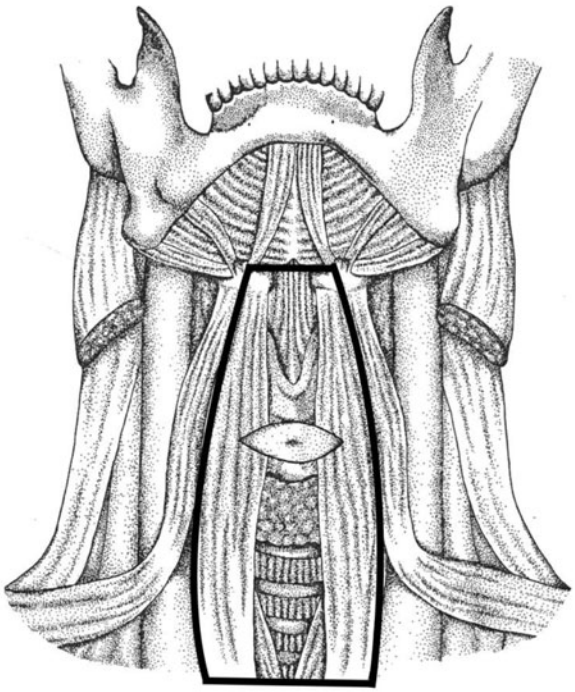

FIG. 2
Extent of the wide anterior neck dissection, including an ellipse of skin. Sternocleidomastoid muscles have been removed to demonstrate area of excision.

encompassed in this dissection. The lateral extent of dissection is the lateral border of the sternohyoid muscle.

If not already excised from a previous procedure, the middle 3 cm of the hyoid bone is resected. Figure 2 depicts the area of resection. Thereafter, an assistant applies downward pressure onto the tongue through the oral cavity, and a wedge-shaped core of tissue is resected from the tongue base to encompass the foramen caecum. The specimen is removed en bloc; if a sinus tract is present, the specimen should include the ellipse of incised skin with attachment maintained to the underlying platysma and fascia. No attempt should be made to skeletonise the duct as this may preclude removal of any ductules.

Figure 3 illustrates the neck after wide anterior neck dissection. A drain is placed and the neck is closed in two layers, taking care to avoid excessive tension at the site of closure.

Results

Between 1 January 2009 and 1 January 2015, seven adults (14.3 per cent female; mean age, 26.4 ± 10.9 years) underwent wide anterior neck dissection for recurrent thyroglossal duct cysts or sinuses. Patient and surgical details are outlined in Table I.

The primary management method was cyst aspiration, followed by cystectomy in three patients, the Sistrunk procedure in three patients, and aspiration with a subsequent Sistrunk operation in one patient.

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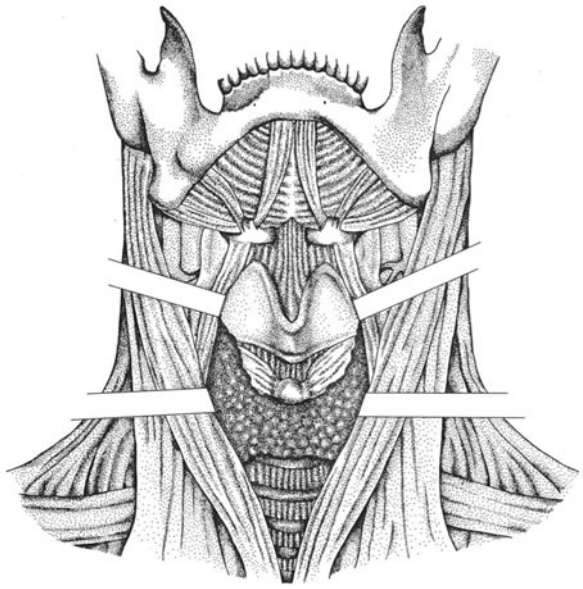

FIG. 3
Anterior neck after wide dissection demonstrated through retraction of the sternocleidomastoid and omohyoid muscles.

Average time to recurrence was 18 ± 9.8 months. Presentation entailed recurrence of the thyroglossal duct cyst (57 per cent) or sinus tract (43 per cent). All three patients presenting with recurrent sinus tracts had cutaneous involvement prior to their primary procedure.

Following wide anterior neck dissection, histopathological analysis confirmed the presence of a remnant thyroglossal duct in all cases.

Post-operative complications included seroma, in two cases, and an isolated case of wound necrosis, dehiscence and cellulitis requiring antibiotics and specialist wound care. Each patient was followed up for 12 months, during which time no thyroglossal duct cysts or sinuses recurred.

Discussion

There is no consensus regarding the management of recurrent thyroglossal duct cysts and sinuses in the adult population. Given the paucity of literature, therapeutic considerations surrounding the extent of surgical resection for this predominantly benign disease process continue to be deliberated.

Recurrence is attributed to incomplete excision, particularly in the hyoid and suprahyoid localities, but may also result from failure to account for infrahyoid ductule branching.[9,17,18] Sade and Rosen histologically reviewed 20 en bloc excisions of thyroglossal duct cysts, and reported that 14 patients had multiple ducts that extended from the hyoid bone to the foramen caecum.[19] Horisawa *et al*. reconstructed 10 thyroglossal duct tracts and demonstrated significant infrahyoid branching, with ductules merging into a single duct at the hyoid bone, anteriorly and posteriorly.[10] These histopathological findings indicate the need for surgical considerations to address the significant embryological variability related to thyroglossal duct cysts and sinuses.

Thyroglossal duct cyst aspiration, or incision and drainage, provide immediate symptomatic relief, but are greatly associated with recurrence (in over 50 per cent of cases) and are not recommended for definitive management.[7,9] Furthermore, given the intimate relationship of the thyroglossal duct to the hyoid bone, management that does not involve removal of the central aspect of the hyoid increases the risk of recurrence.[9] In our series, three patients did not have their hyoid bone removed as part of their primary surgery and subsequently experienced recurrence. The remaining cases of recurrence can be attributed to the failure of primary resection to encompass all thyroglossal duct branches.

The anatomical variability of thyroglossal duct cysts and sinuses should be addressed when managing recurrences. A Sistrunk procedure may not completely excise all arborising ducts given the limited anterior excision.[17] However, in view of the low recurrence and morbidity associated with the Sistrunk procedure, the authors do not support a wider excision for primary disease.

Mickel and Calcaterra were the first to recognise the importance of excising all tracts, and performed wide anterior neck dissection on four patients with recurrent thyroglossal duct cysts.[8] Individual case analyses were

TABLE I
PATIENT, PROCEDURE AND FOLLOW-UP DATA

Pt no.	Sex, age (years)	Primary intervention		Recurrence		Wide anterior neck dissection	
		Cyst aspiration?	Operative intervention	Presentation	Time (months)	Follow up (months)	Further recurrence?
1	M, 34	Yes	Sistrunk	Thyroglossal duct cyst	10	12	No
2	M, 48	Yes	Cystectomy	Thyroglossal duct cyst, cellulitis	28	12	No
3	M, 21	Yes	Cystectomy	Sinus, cellulitis	30	12	No
4	M, 18	No	Sistrunk	Sinus, cellulitis	6	12	No
5	F, 21	No	Sistrunk	Thyroglossal duct cyst	11	12	No
6	M, 25	Yes	Cystectomy	Thyroglossal duct cyst	26	12	No
7	M, 18	No	Sistrunk	Sinus, cellulitis	15	12	No

Pt no. = patient number; M = male; F = female

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not described; however, the authors noted no recurrences between 1 month and 20 years of follow up. Thereafter, Howard and Lund described a similar en bloc surgical strategy for primary resection in 14 thyroglossal duct cyst patients.[20] Again, individual patient data were not provided; however, no recurrences were reported in the cohort. Finally, Patel *et al*. presented 2 cases of recurrent thyroglossal duct cysts in adults successfully managed with wide anterior local excision, and reported no recurrences over a 9.5-month average follow-up period.[18] In comparison to our surgical technique, Patel and colleagues' dissection was limited laterally to a narrow aspect of the central strap muscles and inferiorly to the border of the thyroid isthmus. Given the variability in the location of thyroglossal duct cysts and sinuses, we undertake a wider dissection for managing recurrence. In our series of 7 adult patients, we removed arborising ducts with a wide anterior neck dissection and are yet to experience a recurrence over an average 12-month follow up period.

- **Thyroglossal duct cyst or sinus occurrence and recurrence in adults is very rare**
- **Recurrence after primary resection is attributed to anatomical variability and retained thyroglossal ducts**
- **Wide anterior neck dissection should be considered for the management of recurrences in adults**

The extensive fascial and muscular resection of the wide anterior neck dissection is not without significant risk and potential morbidity.[18] Patel *et al*. described one patient requiring a temporary tracheostomy following a post-operative haematoma.[18] In addition, Perkins *et al*. reported scarring and localised dermatitis as long-term sequelae in children.[21] In our cohort, two of the earlier patients experienced minor post-operative seromas that were successfully managed with needle aspiration. Consequently, we modified our technique and recommend routine drain placement to minimise this occurrence. Our practice is to remove the drain when the output is less than 20 ml over 24 hours, which in our experience is usually post-operative day 2. Tension at the site of skin closure is of additional concern, particularly when wide elliptical excision of the skin has been performed around a recurrent cutaneous sinus. Indeed, we report one case of wound necrosis, dehiscence and cellulitis requiring intravenous antibiotics and daily dressings.

Conclusion

Appropriate management of recurrent thyroglossal duct cysts and sinuses in adults is underreported in the literature. Based on the available data and the seven cases described in this study, we advocate wide anterior neck dissection for the patients affected by this disease.

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References

1 Al-Khateeb TH, Al Zoubi F. Congenital neck masses: a descriptive retrospective study of 252 cases. *J Oral Maxillofac Surg* 2007;**65**:2242–7
2 Ellis PD, van Nostrand AW. The applied anatomy of thyroglossal tract remnants. *Laryngoscope* 1977;**87**:765–70
3 Kurt A, Ortug C, Aydar Y, Ortug G. An incidence study on thyroglossal duct cysts in adults. *Saudi Med J* 2007;**28**:593–7
4 Ducic Y, Chou S, Drkulec J, Ouellette H, Lamothe A. Recurrent thyroglossal duct cysts: a clinical and pathologic analysis. *Int J Pediatr Otorhinolaryngol* 1998;**44**:47–50
5 Allard RH. The thyroglossal cyst. *Head Neck Surg* 1982;**5**:134–46
6 Foley DS, Fallat ME. Thyroglossal duct and other congenital midline cervical anomalies. *Semin Pediatr Surg* 2006;**15**:70–5
7 Oomen KP, Modi VK, Maddalozzo J. Thyroglossal duct cyst and ectopic thyroid: surgical management. *Otolaryngol Clin North Am* 2015;**48**:15–27
8 Mickel RA, Calcaterra TC. Management of recurrent thyroglossal duct cysts. *Arch Otolaryngol* 1983;**109**:34–6
9 Pollock WF, Stevenson EO. Cysts and sinuses of the thyroglossal duct. *Am J Surg* 1966;**112**:225–32
10 Horisawa M, Niinomi N, Ito T. Anatomical reconstruction of the thyroglossal duct. *J Pediatr Surg* 1991;**26**:766–9
11 LaRiviere CA, Waldhausen JH. Congenital cervical cysts, sinuses, and fistulae in pediatric surgery. *Surg Clin North Am* 2012;**92**:583–97
12 Schlange H. About the congenital neck fistula [in German]. *Arch Klin Chir* 1893;**46**:390–2
13 Sistrunk WE. Technique of removal of cysts and sinuses of the thyroglossal duct. *Surg Gynaecol Obstet* 1928;**46**:109–12
14 Galluzzi F, Pignataro L, Gaini RM, Hartley B, Garavello W. Risk of recurrence in children operated for thyroglossal duct cysts: a systematic review. *J Pediatr Surg* 2013;**48**:222–7
15 Hoffman MA, Schuster SR. Thyroglossal duct remnants in infants and children: reevaluation of histopathology and methods for resection. *Ann Otol Rhinol Laryngol* 1988;**97**:483–6
16 Hirshoren N, Neuman T, Udassin R, Elidan J, Weinberger JM. The imperative of the Sistrunk operation: review of 160 thyroglossal tract remnant operations. *Otolaryngol Head Neck Surg* 2009;**140**:338–42
17 Kim MK, Pawel BR, Isaacson G. Central neck dissection for the treatment of recurrent thyroglossal duct cysts in childhood. *Otolaryngol Head Neck Surg* 1999;**121**:543–7
18 Patel NN, Hartley BE, Howard DJ. Management of thyroglossal tract disease after failed Sistrunk's procedure. *J Laryngol Otol* 2003;**117**:710–12
19 Sade J, Rosen G. Thyroglossal cysts and tracts. A histological and histochemical study. *Ann Otol Rhinol Laryngol* 1968;**77**:139–45
20 Howard DJ, Lund VJ. Thyroglossal ducts, cysts and sinuses: a recurrent problem. *Ann R Coll Surg Engl* 1986;**68**:137–8
21 Perkins JA, Inglis AF, Sie KC, Manning SC. Recurrent thyroglossal duct cysts: a 23-year experience and a new method for management. *Ann Otol Rhinol Laryngol* 2006;**115**:850–6

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