

Original Article

Management of thyroglossal duct cysts in children

ZAFER TÜRKYILMAZ,¹ KAAAN SÖNMEZ,¹ RAMAZAN KARABULUT,¹
BILLUR DEMİRĞOULLARI,¹ CEM SEZER,² A. CAN BAŞAKLAR¹ AND NURI KALE¹

Departments of ¹Pediatric Surgery and ²Pathology, Gazi University Medical Faculty, Ankara, Turkey

Abstract

Background: The recurrence rate of thyroglossal duct cysts removed by Sistrunk or other procedures is 4% and 50%, respectively. The aim of the present study was to explain the reasons for recurrence and misdiagnosis.

Methods: Twenty-seven children underwent surgery for thyroglossal cysts and fistulas during 1989–2000. Age, sex, length of history, presentation, preoperative investigations, operative findings, histopathology of the lesion and length of excised hyoid bone, postoperative complications and length of follow-up were recorded. Statistical analysis was performed using Fisher's exact test with a significance level of $P < 0.05$.

Results: Twenty-three patients were primary referrals and four were secondary referrals having had previous surgery with misdiagnosis. The recurrence rate after a Sistrunk procedure was similar to the rate indicated in the literature (3.7%); however, no related special features could be identified such as inflammation ($n = 12$), perforation at surgery ($n = 7$), presentation with fistula ($n = 4$) or previous drainage of abscess ($n = 5$) ($P > 0.05$).

Conclusion: Misdiagnosis is the most common cause of inadequate and inappropriate surgery, leading to recurrence of the lesion. The authors recommend a Sistrunk procedure for all cases of suspected thyroglossal duct cysts.

Key words

children, Sistrunk procedure, surgery, thyroglossal duct cyst.

Thyroglossal duct cysts (TDC) have been considered to be the most common congenital midline neck swelling in children, but the incidences of thyroglossal and branchial anomalies are approximately equal in a recent large pediatric series.¹ While TDC are usually recognized by the age of 5 years, at least half of the lesions are diagnosed in the second decade of life.^{1,2} The majority of TDC lie close to the hyoid bone, however, they can be at any site along the pathway of descent of the thyroid anlage.³ TDCs most often present with a painless swelling, a draining sinus or a tender mass.^{1,4} Movement of the cyst with swallowing is often cited as a reliable diagnostic sign but can be difficult to evaluate in young children and misdiagnosis could occur due to the similar manifestation of the dermoid cyst, lymphadenopathy and cystic hygroma when located near the hyoid bone.^{5,6} Unusual locations of TDC have also been described.^{7–9} Embryologic development of neck structures is a reason that these remnants can be found in other locations.⁷

Attention has been drawn to the fact that the duct is frequently multiple and branched.^{10,11} TDC and sinuses are usually lined by respiratory or non-keratinizing stratified squamous epithelium or both.^{11,12}

Excision of the hyoid body and cyst was first proposed by Schlang in 1893 (recurrence rate 20%) and Sistrunk popularized the procedure in 1920, advising removal of a tissue core from the hyoid bone to the foramen cecum, reducing the recurrence rate to 3%.^{3,4}

Despite the large body of literature supporting Sistrunk's approach, many cases of TDC continue to be misdiagnosed and mismanaged with resulting recurrences. The purpose of the present study is to report our long-term results with Sistrunk's procedure for TDC to determine the cause of recurrences.

Methods

Hospital records of children operated for TDC or sinuses in the period February 1989 to May 2000 at Department of Pediatric Surgery, Gazi Medical Faculty, Gazi University, Ankara, Turkey, were reviewed. Age, sex, length of history,

presentation, preoperative investigations, operative findings, histopathology of the lesion and length of excised hyoid bone, postoperative complications and follow-up evaluations with particular emphasis on recurrences were noted.

Preoperative evaluation consisted of physical examination with regard to movement of the mass with protrusion of the tongue, ultrasonographic assessment of the mass or draining sinuses in all of the patients and radioactive iodine isotope scans in nine patients.

Routine histopathological sections were re-evaluated. Diagnostic criteria were based on the presence of ductular and/or cystic structures lined by cuboidal or columnar (respiratory) epithelium or non-keratinizing stratified epithelium without epidermal appendages. The length of excised hyoid bone was also measured. Inflammatory findings were noted if present.

Statistical analysis was performed using Fisher's exact test with a significance level of $P < 0.05$.

Results

The mean age of the 27 patients was 6.25 years, ranging between 18 months and 14 years with 18 patients younger than 7 years. There were 15 boys and 12 girls (1.25 male preponderance). Twenty-three patients were primary referrals and four were secondary referrals having had previous surgery with misdiagnosis.

The duration of symptoms before admission ranged from 20 days to 6 years and averaged 16 months.

Eighteen patients (66%) were admitted with midline cystic mass, five cases (18.5%) with typical glairy discharge, and four patients (14.8%) with serious cervical infection requiring drainage and antimicrobial therapy. Two of these cases with infection were followed for deep cervical infection in other clinics and diagnosed as having TDC after resolution of the infection. The cases with draining sinuses had no history of previous infection and no documented bacterial growth.

The lesions were localized in the midline at the hyoid level in 22 (81%), infrahyoidal level in four (one localized slightly to left of midline) and the suprahyoidal level in one of the patients.

In none of the four infected cases drained previously did a fistula develop before operation. Two of these children had a history of upper respiratory tract infection a short time before infection of the lesions occurred.

Ultrasonographic examinations were consistent with anechoic pattern in seven (31.8%), pseudosolid in nine (40.9%) and heterogenous in six (27.2%) cases. Although cystic lesions were not documented in cases with fistula, ultrasonographic examination was nevertheless performed in

order to assess the thyroid gland. In all cases, ultrasonography revealed normal thyroid localization and patterns in this series. Thyroid scintigraphy was done in nine patients to document whether ectopic thyroidal tissue was present, however, it was eliminated later because of the success of ultrasonography.

Twenty-three patients were treated by single operations using Sistrunk's approach with dissection of the cyst or fistulous tract, excision of part of the hyoid bone centrally and coring out muscle attached to the hyoid bone to a level near foramen cecum. In no case was the mouth entered. Four cases required Sistrunk's procedure as a second procedure for cure. Two of these children had simple excision of their cysts with misdiagnosis of dermoid cyst in other adult surgical clinics. Another two children were operated previously in our clinic, one with a simple cyst excision for a misdiagnosis of fistulized submental lymphadenitis and one with incomplete Sistrunk's procedure, following the suprahyoid fistula duct, instead of coring out the muscular structures.

The number of complicated cases was 12 (44%), including four patients with infected cysts requiring drainage preoperatively, five with fistula and three with purulent cystic discharge encountered during the operations. Operative documents revealed inadvertent cyst rupture in five (18.5%) and disruption of suprahyoidal tract in two cases (7%).

The wound was drained in 11 cases for 24–48 h. Postoperative complications consisted of hematoma development in one patient and seroma collection in another patient. Patients with previous infections, those with peroperative purulent discharge and those with wound drainage received antibiotics.

Histopathological evaluation revealed findings consistent with the presence of thyroglossal remnant in all cases. The cystic and/or ductular epithelium was ciliated in seven, non-keratinizing in 11 and mixed in nine specimens. Inflammatory changes were severe in six cases and less severe in seven cases. Multiple ductular structures were found in 23 specimens (85%). The lengths of resected hyoid bones ranged from 9 mm to 21 mm (average 14.5 mm). No ectopic thyroidal tissue was seen.

Follow-up periods ranged from 2 to 13 years with an average of 5 years. Recurrence was seen in only one case (3.7%) following a Sistrunk's procedure, 3 years after the procedure. Other recurrences were associated with simple cyst excisions due to misdiagnoses after 6 months in two cases and 1 year in one case.

The rate of recurrence was not statistically different between cases with peroperative cystic rupture or ductal severement (1/7) and those cases without rupture (0/20) ($P > 0.05$). The rate of recurrence was also insignificant between cases with infection or fistula (0/9) and those without infection or fistula (1/18) ($P > 0.05$).

Discussion

Thyroglossal duct cysts usually present as asymptomatic midline cervical masses. However, due to a considerable infection rate, reported to be as high as 50% in the medical literature, and to presence of other midline cervical masses such as dermoid cysts, misdiagnosis and consequent mismanagement can ensue.^{3,4,13,14} A not so low number of mismanaged cases due to an initial misdiagnosis in this series reaching an incidence of nearly 20% emphasizes the importance of a correct diagnosis in the first place. It should be remembered that dermoid cysts, second in incidence to TDC, are reported to sometimes occur simultaneously with TDC.^{3,7}

Preoperative diagnosis could also emerge as a problem in TDC with unusual localizations.^{7,12,13} We consider the case with an initial diagnosis of submental draining lymphadenitis as a good example of this problem. In a large series, 26 lesions (8%) out of 300 were at the suprahyoidal level.¹² In our series, only one (3.7%) lesion was localized at that position and the presence of a submental draining fistula rather than a cyst brought about a diagnostic dilemma.

It is considered that movement of the mass with protrusion of the tongue is an unreliable diagnostic sign especially in children.¹ Limitation in the movement of the cyst with protrusion of the tongue due to its attachment to foramen cecum has been suggested as a more reliable sign.⁵

We consider ultrasonographic examination to be valuable and sufficient, mainly in the assessment of the thyroid gland. In none of our cases did we encounter an unexpected ectopic thyroid tissue with a normal ultrasonographic thyroid image. This finding should make scintigraphic examination unnecessary. However, the sonographic images of the cysts were variable. A solid pattern referred to as 'pseudosolid' in literature was reported in nine of 22 cases (40.9%), excluding the ones with fistula. This number is similar to the reported incidence of 56.5% in another series.¹⁵ Such a sonographic image could direct one to a diagnosis of a malignant tumor in cases with hard cysts.³

The procedure popularized by Sistrunk has been the most commonly used in the treatment of TDC, with very low recurrence rates.¹² This rate was 3% in Sistrunk's first series as reported by Bennett *et al.*⁴ Recurrence was acceptable at 3.7% in this series. The factors held responsible for recurrences comprised variable entities such as dermal involvement, young patient age (childhood), rupture of the cyst during the operation, lobulation of the cyst, inflammation and/or infection, elimination of the coring-out procedure in an attempt to follow the suprahyoidal tract, and cases with fistulas.^{14,16} Ein *et al.* had considered infection and fistulas as being responsible for recurrence in their large series of 270 cases.¹³ Solomon and Rangecroft, in contrast, had suggested inappropriate surgical approach due to initial misdiagnosis

rather than infection and/or inflammation as the reason for recurrence.¹² In the present series the recurrence rates were the same for fistulous and non-fistulous cases; for those with peroperative cystic rupture and those without; the cases associated with histopathologically proven inflammation and those not associated with it ($P > 0.05$ for each comparison). Preoperative infection also did not affect the recurrence rate in this series. The factor that had the greatest impact for recurrence was incomplete surgical procedure (e.g. simple cyst excision in three and elimination of the coring-out procedure out in one patient) due to misdiagnosis either by our clinic or by other clinics or to mismanagement. We do not consider the patient's young age to be a factor in recurrence if the principles of the operation are strictly adhered to. Those studies that suggest the patient's young age as being a factor in recurrence reported series comprising both adults and children operated in the same clinic.^{6,13,14} We believe that the recurrence rate should decline if childhood operations are performed in pediatric surgical units expertised in managing children.

The oral cavity was not entered in the cases in this series and the core-out procedure fell short of (though very near to) reaching exactly the level of the foramen cecum. We do not have the necessary data to exactly document the length of core-out structures. Although this could be a reflection of Horisawa's suggestion of limiting the core-out procedure in close proximity to the hyoid bone with variations depending on the child's age, we believe that dissecting close to the foramen cecum does not result in higher recurrence rates or morbidity due to branching of the duct into ductuli. It should be noted that the only case in this series that recurred with a correct diagnosis was the one in which the suprahyoidal duct was dissected instead of a core-out procedure being performed. The addition of the coring-out procedure with a wider dissection in the second operation solved the problem. Complications have been minor and limited to the wound in only two patients in the present cases (7%). However, entering the oral cavity is unnecessary as reflected by the very acceptable low recurrence rate in this series. The average length of excised hyoid bones is more than Sistrunk's and closer to Horisawa's suggestions. This resection, we believe, should also be dependent on the age and in turn, the size of the patient. We attempted to resect most of the corpus during the operation.

We feel that a special mention should be made of the dermoid cysts diagnosed during an operation. In our opinion a Sistrunk's procedure should be done for dermoid cysts attached to the hyoid bone either directly with a close proximity to the bone or indirectly. Indeed, DeMello *et al.* have proposed that dermoid cysts of the neck represent variants of TDC with predominating ectodermal elements and advocated Sistrunk's procedure for all dermoid cysts attached to the hyoid bone,¹⁷ which we are in agreement with.

There are also reports describing coexistence of these two lesions.^{3,8} Therefore, a Sistrunk's operation can be justified whenever diagnosis is questioned.

We conclude that a Sistrunk's operation results in a low recurrence rate and we suggest that misdiagnosis is the major factor resulting in an incomplete procedure and high recurrence.

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