

## ORIGINAL ARTICLE

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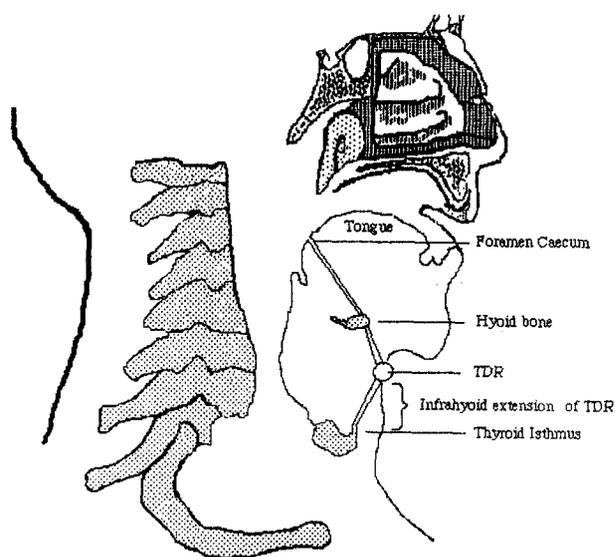
## Thyroglossal duct remnants

### Infrahyoid extension as a cause of recurrence

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**Abstract** Ten children were admitted with recurrences of thyroglossal duct remnants (TDRs) during the years 1978–1993. Unexcised infrahyoid extension of the thyroglossal duct was found to be the cause of recurrence in 3 cases, whereas the remainder were due to misdiagnosis and inappropriate surgery. After surgical treatment, these three cases were evaluated regarding the cause of recurrence. The treatment of choice for these lesions is the Sistrunk procedure. The complete Sistrunk procedure solves the problem in the majority of patients, but in cases with infrahyoid extension tracing of the extension is required. It is concluded that an appropriately performed Sistrunk procedure assures a complete cure in the vast majority of cases of TDRs; however, routine exploration for and excision of the occasional infrahyoid extensions is recommended for the prevention of recurrence.

**Key words** Thyroglossal duct remnant · Sistrunk procedure · Infrahyoid extension



**Fig. 1** Schematic diagram of infrahyoid extensions (TDR thyroglossal duct remnant)

### Introduction

The thyroglossal duct (TD) is a remnant of the diverticulum where the thyroid gland starts to develop from the foramen caecum in the 3–4-week embryo. Descending inferiorly in the midline and crossing the hyoid bone, it reaches its final position by the end of the 7th week [2, 7, 9]. A thyroglossal duct remnant (TDR) is seen as a cyst or sinus opening in the midline at the hyoid region in over 50% of cases, but it may occasionally extend down to the infrahyoid region, communicating with the thyroid isthmus (Fig. 1) [5, 7, 8, 10]. The treatment of choice for these lesions is the Sistrunk procedure, however, there is still a recurrence rate of

3%–5% [1–6, 8–11]. In this study, we report three cases out of ten recurrent TDRs in which rare infrahyoid extensions of the TD were identified as the cause of recurrence.

### Materials and methods

Ten children were admitted with recurrences after TDR operations during the years 1978–1993. Unexcised infrahyoid extension of the TD was found to be the cause of recurrence in three patients, two of them previously operated upon by the authors, whereas the remaining ones were due to misdiagnosis and inappropriate surgery. Other relevant information was obtained from the medical records.

### Case reports

*Case 1.* A 5-year-old male presented with an anterior midline neck mass. A Sistrunk operation was performed by the authors after the

diagnosis of a TD cyst was made. He was readmitted with a recurrence and infectious discharge in the 3rd postoperative year. Exploration revealed an infrahyoid extension as the source of discharge, which was completely excised. The patient is now 16 years old without any further problems.

*Case 2.* A 10-year-old male presented with recurrent infectious discharge from the anterior midline of the neck. He had had a symptomatic TD fistula at the age of 3 years and had been operated upon three times at a local hospital at 5, 7, and 8 years of age. None of these were Sistrunk operations, but were presumably simple cystectomy and subsequent curettage procedures. Infrahyoid extension of a TDR was detected during surgery and a complete Sistrunk operation with removal of the midportion of the hyoid bone, including the infrahyoid extension, was performed by the authors. The patient is still symptom-free after 6 years' follow-up.

*Case 3.* A 5-year-old male presented with recurrent discharge from an anterior midline neck lesion. A cystectomy had been performed 2 years previously followed by a curettage for continuing discharge. A complete Sistrunk procedure was performed by the authors. After admission for a recurrence, an infrahyoid extension of the TD was identified and excised. After 6 years the patient is still symptom-free.

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## Discussion

Certain important pitfalls in the management of TDRs can lead to recurrence, misdiagnosis being the most common cause [1–6, 8–11]. Differential diagnosis of midline neck masses such as bronchial cleft remnants, lipomas, dermoid cysts, lymphadenopathy, hemangiomas, and thyroid masses can usually be done without difficulty [6, 10]. Either a preoperative sinogram or intraoperative dye injection into the sinus is used to delineate the tract [9, 10].

Misdiagnosis leads to incomplete and inappropriate operations. Recurrences of 50%–82% have been reported after simple excision of TDRs [1–6, 8–10]. Simple cystectomy, excision, or curettage was performed in eight of our ten recurrent cases, most of which were operated upon by surgeons who missed this congenital anomalous tract extending to the pharynx. In 1920 Sistrunk stressed two basic points: the excision of the central part of the hyoid bone, and the removal of the proximal and deeper part of the duct. Ein et al. [5], Soucy and Penning [11], and Howard and Lun [8] have stressed the branching and multiplicity of TDRs through the throat as a cause of recurrence despite the Sistrunk procedure. Infection is another important factor: since the tract opens into the throat, bacterial contamination is likely. Infection makes

the ductal tract difficult to locate during excision, and therefore, all infected and necrotic material should be removed [1, 8, 9].

Recurrences despite complete Sistrunk operations in two of our own cases impelled us to search for distal extension of the thyroglossal tract as a cause of recurrence. There were 64 cases of infrahyoid extension among 300 cases reviewed by Solomon and Rangecroft [9]. However, they and other authors have not emphasized the role of infrahyoid extension as a cause of recurrence [5, 8, 11]. The complete Sistrunk procedure solves the problem in the majority of patients, but several other factors may also lead to recurrence.

In conclusion, the complete Sistrunk procedure should be performed for TDRs, however, routine exploration for and excision of the occasional infrahyoid extensions is recommended for the prevention of recurrence after a Sistrunk procedure.

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