

CASE REPORT

Randy L. Hanzlick,¹ M.D.

Lingual Thyroglossal Duct Cyst Causing Death in a Four-Week-Old Infant

REFERENCE: Hanzlick, R. L. "Lingual Thyroglossal Duct Cyst Causing Death in a Four-Week-Old Infant," *Journal of Forensic Sciences*, JFSCA, Vol. 29, No. 1, Jan. 1984, pp. 345-348.

ABSTRACT: A 12-mm spherical thyroglossal duct cyst at the base of the tongue caused the death of a four-week-old infant by obstructing the glottis. Previous case reports of this rare entity indicate that the supine position aggravates airway obstruction. Proof of a cause and effect relationship between such a cyst and death is difficult in the sudden infant death syndrome (SIDS) age group because of nonspecific findings associated with asphyxia and SIDS. Autopsy should include a thorough examination of the oral cavity to detect such lesions.

KEYWORDS: pathology and biology, cysts, asphyxia, airway obstruction, thyroglossal cysts, lingual masses

The author presents the case details of death in a four-week-old infant caused by a lingual thyroglossal duct cyst. Characteristics of this entity and postmortem diagnostic considerations are discussed.

Case Report

A four-week-old black female infant was put to bed in her bassinet at 11 p.m. At 4 a.m. the father found the infant dead in the bassinet in the *supine* position.

The infant was the third child born to a 26-year-old gravida 3, para 3, abortus 0 black female. The previous children were healthy and had uncomplicated pregnancies and deliveries. The only significant prenatal, perinatal, or postnatal history pertaining to the decedent was an episode of aspiration at birth, which resulted in no significant sequelae.

Autopsy demonstrated a well developed female infant with visceral congestion, partially atelectatic lungs, and a few scant petechial hemorrhages on the epicardium and pleura, both anteriorly and posteriorly.

In addition, at the base of the tongue (at the foramen cecum), was a spherical 12-mm cyst with a normal overlying lingual mucosa. With the tongue in the normal retracted position, the cyst rested on the epiglottis and glottis in a "ball valve" fashion (Fig. 1).

The cyst contained a gray-tan mucoid fluid. Grossly, a firm cord extended from the base of the cyst caudad and anterior through the base of the tongue toward the hyoid bone (Fig. 2). The thyroid gland was of normal size, position, and morphology.

Received for publication 23 April 1983; accepted for publication 7 June 1983.

¹Deputy medical examiner, Fulton County, Atlanta, GA.

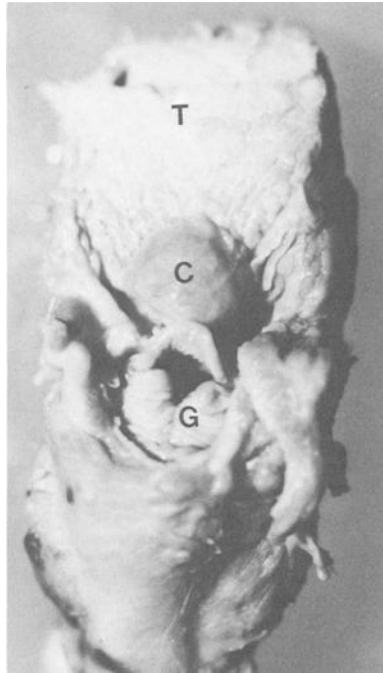


FIG. 1—*Posterior view of tongue and larynx showing cyst (C) resting on epiglottis/glottis (G) at base of tongue (T).*

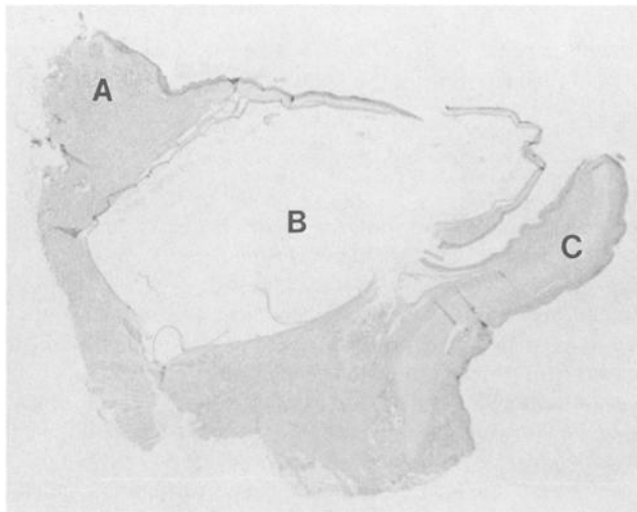


FIG. 2—*Tissue section of cyst (B) showing its relationship to base of tongue (A) and epiglottis (C).*

Histologically, the cyst was lined with a flattened epithelium with squamous characteristics (Fig. 3). No thyroid elements were seen in the cyst wall or surrounding tissues. Sections of the firm area in the tongue and hyoid area showed descent of epithelial elements through the tongue toward the hyoid.

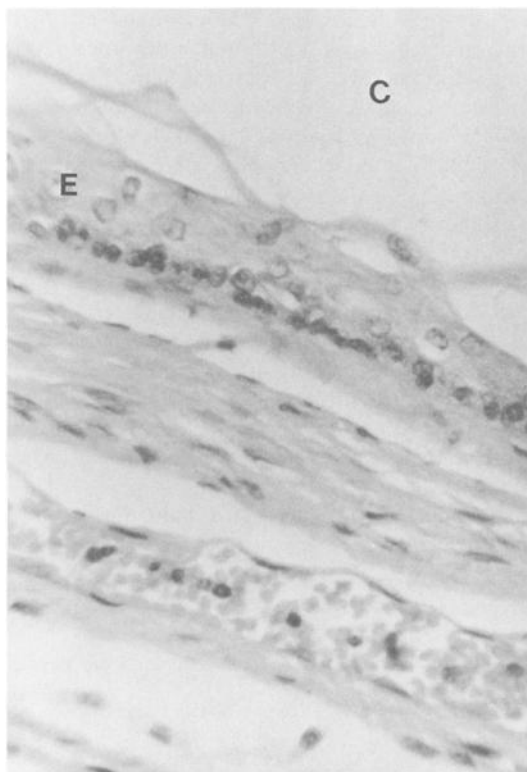


FIG. 3—Tissue section of cyst wall showing squamoid epithelium (E) adjacent to cyst cavity (C).

Discussion

Thyroglossal duct cysts result from a persistence of the embryonic thyroglossal duct. During the fourth week of embryonic development, the thyroid arises as a diverticulum in the pharyngeal floor between the first pharyngeal pouches. The diverticulum then descends caudad and anterior in the neck, remaining attached to the pharyngeal floor at the site that later becomes the foramen cecum of the dorsal tongue surface. By the sixth week of embryonic development, the resulting thyroglossal duct usually becomes a solid stalk, then degenerates. If degeneration does not occur, thyroglossal cysts may subsequently develop anywhere along the path of the persistent thyroglossal duct [1,2].

Thyroglossal duct cysts usually occur below the level of the hyoid, while solid abnormalities are much more frequent above the hyoid within the tongue [1]. However, thyroglossal cysts may occur on the dorsal aspect of the tongue [3]. The differential diagnosis includes dermoid cysts, cervical lymphadenopathy, lipoma, hemangioma, and ectopic or "lingual" thyroid [4]. The outstanding gross feature of thyroglossal cysts is their midline location. Grossly, the thyroglossal cysts usually contain a tan-gray mucoid fluid. Microscopically, the cysts may be lined with transitional, cuboidal, columnar, squamous, ciliated, or nonciliated epithelium [1,2]. Thyroid epithelium is seen in less than 5% of cases [1].

Most cases of thyroglossal cysts manifest themselves by age five, with many cases presenting in the first year of life. They are usually discovered because of disfigurement in the neck or recurring inflammatory disease [2].

However, there are at least two reported cases of lingual thyroglossal cysts presenting with

dyspnea during the first six weeks of life; one of these proved fatal [3]. These two patients developed dyspnea in the supine position which cleared in the prone position. It is interesting, if not significant, that the subject of this case report was found in the supine position.

Investigation of dorsal lingual masses should include a thorough examination of the thyroid gland. In lingual thyroid, a normal thyroid gland is usually absent [5]. Sections of the base of the tongue and hyoid should be examined for epithelial tracts; their presence supports the diagnosis of thyroglossal cysts.

It is also worthy of mention that thyroglossal duct remnants may result in cysts which contain disorders seen in the thyroid gland, such as carcinoma, adenoma, thyroiditis, and thyrotoxicosis [6-9].

It could be argued that death of the infant presented in this paper was a result of sudden infant death syndrome (SIDS), and the thyroglossal cyst was an incidental finding. However, many of the factors commonly associated with SIDS victims were absent, and the age of the infant was not typical of SIDS [10, 11]. Furthermore, previous case reports of documented dyspnea caused by similar lesions, the supine position of the decedent, and the demonstrable "ball valve" effect in this case strongly support the diagnosis of airway obstruction as a result of a lingual thyroglossal cyst.

Autopsy examination should include a thorough examination of the oral cavity and pharynx to detect and evaluate such lesions.

References

- [1] Pollack, W. F. and Stevenson, E. O., "Cysts and Sinuses of the Thyroglossal Duct," *American Journal of Surgery*, Vol. 112, Aug. 1966, pp. 225-231.
- [2] Gross, R. E. and Connerley, M. L., "Thyroglossal Cysts and Sinuses," *New England Journal of Medicine*, Vol. 223, No. 16, 17 Oct. 1940, pp. 616-624.
- [3] Lewison, M. M. and Lim, D. T., "Apnea in the Supine Position as an Alerting Symptom of Tumor at the Base of the Tongue in Small Infants," *Journal of Pediatrics*, Vol. 66, No. 6, 1965, pp. 1092-1093.
- [4] Telander, R. L. and Deane, S. A., "Thyroglossal and Branchial Cleft Cysts and Sinuses," *Surgical Clinics of North America*, Vol. 57, No. 4, Aug. 1977, pp. 779-791.
- [5] Sommers, S. C., "Thyroid Gland," in *Pathology*, W. A. D. Anderson and J. M. Kissane, Eds., Mosby, St. Louis, 1977, p. 1629.
- [6] Keeling, J. M. and Ochsner, A., "Carcinoma in Thyroglossal Duct Remnants: Review of Literature and Report of 2 Cases," *Cancer*, Vol. 12, No. 3, May-June 1959, p. 596-600.
- [7] Early, C. M., "Hashimoto's Disease (Struma Lymphomatosa) Occurring in a Thyroglossal Duct Cysts," *American Journal of Surgery*, Vol. 101, 1961, p. 819-823.
- [8] Tanaka, K. and Civia, W. H., "Cancer Arising in Thyroglossal Duct Remnant," *Archives of Surgery*, Vol. 86, March 1963, p. 466-470.
- [9] Bochetto, J. F., Montoya, A., and Sunde, E. A., "Papillary Carcinoma of the Thyroid in Thyroglossal Duct Cysts," *American Journal of Surgery*, Vol. 104, Nov. 1962, p. 773-776.
- [10] Shannon, D. C. and Kelly, D. H., "SIDS and Near-SIDS," *New England Journal of Medicine*, Vol. 306, No. 16, 22 April 1982, pp. 959-963.
- [11] Hanzlick, R. L., Bruner, B., and Stivers, R. R., "SIDS: A Guide for Medical Examiners," Georgia Sudden Infant Death Syndrome Project, Atlanta, GA, 1982.

Address requests for additional information to
Randy L. Hanzlick, M.D.
Deputy Medical Examiner
Fulton County
50 Coca Cola Pl., S.E.
Atlanta, GA 30303