Journal of Current Medical Research and Opinion

Received 01-10-2023 Revised 04-10-2023 Accepted 23-10-2023 Published Online 24-10-2023

DOI: https://doi.org/10.52845/CMRO/2023/6-10-3 ISSN (O) 2589-8779 | (P) 2589-8760

CMRO 06 (10), 1766-1769 (2023)

Clinical Case Reports

Early Postoperative Death in a child Undergoing Thyroglossal dust cysts Surgery

Mohamed Djilali Merzoug¹ | Mohamed Amine Boumelik¹ | Yassine Merad² | Derouicha Matmour² | Zakaria Merad³ | Adel Tahraoui⁴ | Ouziane Megherbi⁴

¹Forensic department, *Hassani Abdelkader* hospital, UDL
²Laboratory department, *Hassani Abdelkader* hospital, UDL University
³Pathology department, *Hassani Abdelkader* hospital, UDL University
⁴ENT department, *Hassani Abdelkader* hospital, UDL
⁴UDL University



Abstract:

Thyroglossal duct cysts is a common congenital cervical disease, its obvious signs include having a small lump or opening on the front of their neck at birth. Rarely, especially if the mass is located at the base of the tongue, airway obstruction and dyspnea can ensue.

A 11-year-old female with no particular pathological history was admitted for surgical removal of a surinfected cyst of the thyroglossal tract. Few hours after being discharged from the department, the patient was readmitted to the emergency room.

The final diagnosis of fatal asphyxia due to compressive hematoma extended to cervical muscles and para laryngotracheal was determined by both autopsy and histology

Thus, it is vital to always consider all the possible injuries, namely thyroid, salivary gland, and tongue

Copyright : © 2023 The Authors. Published by Publisher. This is an open access article under the CC BY-NC-ND license (https://creativecommons.org/licenses/by-nc-nd/4.0/).

Introduction:

Thyroglossal duct cysts are the most common congenital cervical anomaly, they are reported in about 7% of the population worldwide [1]. This pathology is resulting from the failure of thyroglossal duct to involute during embryological development [2].

Thyroglossal duct cysts can form anywhere along the thyroid's route of migration between the tongue and the inferior neck. They often present as midline neck cysts closely associated with the hyoid bone. Usually, this malformation is not painful, though it might be if the area becomes infected. Malignant thyroglossal duct cyst is a rare condition, occurring in 1% of all cases of thyroglossal duct cysts [3].

Less encountered complications have been reported such as dyspnea [4,5,6,7], dysphagia [8].

In addition, thyrolingual cysts are often containing mixed mucus [9], and were associated with sudden death in infancy [10], and relatively large posterior lingual midline cysts were identified at autopsy [11].





Mohamed Djilali Merzoug / Early Postoperative Death in a child Undergoing Thyroglossal dust cysts Surgery

A thyroglossal duct cyst may appear similar to other growths around the neck (e.g., epidermoid cyst, brachial cleft cyst, and bronchogenic cyst) [12,13].

In infants, thyroglossal cyst is a common cause of airway blockage [14,15,10,16,11,17-20]

Dr. Sistrunk recommended removal of not only the cyst and central portion of the hyoid bone, but also a central core of deep tongue musculature. By doing so, the rate of recurrence is decreased from approximately 50 to 3-5% [21].

A laryngotracheal injury is a rare and possibly severe complication of the Sistrunk surgical procedure leading to problems with the airway, swallowing, and/or voice [1].

We describe a case of early postoperative death in a child.

A 11-year-old female with no particular pathological history was admitted in a liberal health facility for surgical removal of a superinfected cyst of the thyroglossal tract.

The surgery was successfully completed under general anesthesia and the patient was then completely discharged. However, few hours later, the child was readmitted for acute respiratory distress, cyanosis, and mottling, followed by an unexpected death.

Autopsy was performed highlighting compressive hematoma extended to the whole of the cervical muscles and laryngotracheal area, associated to significant glottis and laryngeal edema.

Although, thyroglossal cysts might have compressive symptoms of the airway, mortality could have been prevented by meticulous examination to the possible surgical wounds.



Figure 1: postmortem aspect of the neck after surgery



Figure 2: images of postoperative laryngeal edema hematoma

Current Medical Research and Opinion, Vol. 06, Issue. 10, Page no : 1766-1769 DOI : https://doi.org/10.52845/CMRO/2023/6-10-3 Page | 1767

Case report:

Discussion:

By browsing the literature on case reporting we came across other authors who have described various clinical cases of thyroglossal dust cysts, for an instance, a 69-year-old female presented for a neck lump and dysphagia for 1 year [2]. Moreover, a fatal asphyxia by a thyroglossal duct cyst has been described in a healthy 35-year-old man who presented a sudden throat pain associated to an inability to swallow [22]. Furthermore, a 55-year-old man reportedly died from fatal unexpected asphyxia caused by a thyroglossal cysts mass [9].

Unusual presentations are mainly seen among young children and are responsible of death in half of these cases [9]. In children, anterior midline or near midline neck masses may be often ascribed to thyroglossal duct cysts and can lead to respiratory distress [23].

The Sistrunk surgical procedure is widely used for treatment of thyroglossal duct cyst [24].

Cyst removal is safe and well-tolerated procedure in the adult population [25], however, postoperative-y, periodic monitoring with neck CT and/ or u-trasound is important in patients treated with the Sistrunk procedure, given the high incidence of synchronous malignancy especially in adults [24].

Surgery adverse events are typically readmissions, wound infections, and unplanned reoperations [23]. Almost 40% of postoperative deaths occurred within three days of emergency surgery [26].

Reportedly, the presence of thyroglossal duct cyst may cause complications such as upper airway obstruction, and dyspnea [10,1,9,27,14-20]. Moreover, the risk of reoperation and surgical-site reinfection should be considered [25].

Risk factors that may increase the risk of recurrence are multiple duct tracts, surgery during active exacerbation, and inappropriate surgical technique [28.29].

Similarly, a man died from asphyxia within 1 hour after the initial symptoms, autopsy revealed a 1.5 cm ruptured thyroglossal duct cyst with secondary swelling of the epiglottis and tongue, and thus resemble to our findings [22]. Furthermore, laryngotracheal injury is rare and can induce issues with the airway, swallowing, and voice [1]. which is in line with our observation.

Conclusion:

Thyroglossal duct cysts typically present as mobile neck masses that may cause rare fatal airway obstruction among children.

In our case the cause of death was attributed to asphyxia by airway compression secondary to extended laryngeal hematoma following thyroglossal excision. These findings are suggesting a possible mishandled thyroid injury.

References:

- 1. Amos J, Shermetaro C. Thyroglossal Duct Cyst. StatPearls, 2023
- 2. Tachamo N, Le B, Driben J, Magaji V. Locally Advanced Thyroglossal Duct Cyst Carcinoma Presenting as a Neck Mass. Case Reports in Endocrinology, vol. 2017, Article ID 7014313, 4 pages, 2017.
- 3. Gertallah LM, Eltokhy E, Baiomy TA, Harb OA. Invasive primary papillary carcinoma in a thyroglossal duct cyst: A case report and review of the literature, Human Pathology: Case Reports, Volume 15, 2019, Pages 7-12,
- Strachan D, Wengraf C. An unusual thyroglossal cyst causing upper airway obstruction. Br J Clin Pract 1966;50(8):472–3.
- 5. McDonald DM. Thyroglossal cysts and fistulae. Int J Oral Surg 1974;3:342–6.
- Perez-Calderon R, Gonzalo-Garijo MA, Albendiz-Ruiz V. Intermittent stridor and dyspnea in an adult. Ann Allergy Asthma Immunol 2003;90(3):294–7.
- Olumide F. Thyroglossal cyst presenting with severe laryngeal obstruction. Niger Med J 1973;3(1):52–4.
- Lubben B, Alberty J, Lang-Roth R, Seifert E, Stoll W. Thyroglossal duct cyst causing intralaryngeal obstruction. Otolaryngol Head Neck Surg 2001;125(4):426–7.

Mohamed Djilali Merzoug / Early Postoperative Death in a child Undergoing Thyroglossal dust cysts Surgery

- Sauvageau A, Belley-Côté EP, Racette S. Fatal asphyxia by a thyroglossal duct cyst in an adult. J Clin Forensic Med. 2006 Aug-Nov;13(6-8):349-52.
- Byard RW, Bourne AJ, Silver MM. The association of lingual thyroglossal duct remnants with sudden death in infancy. Int J Pediatr Otorhinolaryngol 1990;20:107– 12.
- Hanzlick RL. Lingual thyroglossal duct cyst causing death in a fourweek-old infant. J Forensic Sci 1984;29(1):345–8.
- 12. Merad Y, Derrar H, Zeggai A, Chadli M, Bemrah N, ElHabachi B. Primary splenic hydatid cyst an unexpected diagnosis: Case report. Annals of Medicine and Surgery 65, 102293, 2021
- 13. Merad Y, Derrar H, Zeggai A, Belkacemi M, Belmokhtar Z, Adjmi-Hamoudi H. A rare primary hydatid cyst of the psoas muscle in a rural setting: A case presentation. Annals of Medicine and Surgery 59, 86-88, 2020
- Lewison MM, Lim DT. Apnea in the supine position as an alerting symptom of tumor at the base of the tongue in small infants. J Pediatr 1965;66(6):1092–3.
- 15. Paez P, Warren WS, Srouji MN. Stridor as the presenting symptom of lingual thyroglossal duct cyst in an infant. Clin Pediatr 1974;13:1077–8.
- Solomon JR, Rangecroft L. Thyroglossal duct lesion in childhood. J Pediatr Surg 1984;19:555–60.
- Kuint J, Horowitz Z, Kugel C, Toper L, Birenbaum E, Linder N. Laryngeal obstruction caused by lingual thyroglossal duct cyst presenting at birth. Am J Perinatol 1997;14(6):353–6.
- Samuel M, Freeman NV, Sajwany MJ. Lingual thyroglossal duct cyst presenting in infancy. J Pediatr Surg 1993;28:891–3.
- Mahboudi S, Gheyi V. MR imaging of airway obstruction in infants and children. Int J Pediatr Otorhinolaryngol 2001;27:219–27.
- 20. Weldon BC, Krafcik JM. Breath-holdinglike spells in an infant: an unusual

presentation of lingual thyroglossal duct cyst. J Pediatr Surg 2000;35(9):1381–4.

- Gallagher TQ, Hartnick CJ. Thyroglossal duct cyst excision. Adv Otorhinolaryngol. 2012;73:66-9.
- 22. Kay HE. Fatal thyroglossal cyst. J Laryngol Otol 1956;70(4):240–1
- 23. Cheng J, Lerebours R, Lee HJ. Current trends and 30-day surgical outcomes for thyroglossal duct cyst excision in children. Int J Pediatr Otorhinolaryngol. 2020 Jan;128:109725.
- 24. Ashima Mittal, Ariel Sandhu, Mamta Chhetri, Gayatri Jaiswal, A Rare Case of Thyroglossal Duct Cyst Cancer and Literature Review, JCEM Case Reports, Volume 1, Issue 2, March 2023, luad036,
- Anderson JL, Vu K, Haidar YM, Kuan EC, Tjoa T. Risks and complications of thyroglossal duct cyst removal. Laryngoscope. 2020 Feb;130(2):381-384.
- 26. Aggarwal G, Broughton KJ, Williams LJ, Peden CJ, Quiney N. Early Postoperative Death in Patients Undergoing Emergency High-Risk Surgery: Towards a Better Understanding of Patients for Whom Surgery May Not Be Beneficial. J Clin Med. 2020 Apr 29;9(5):1288.
- 27. Ogunkeyede SA, Ogundoyin OO. Management outcome of thyroglossal cyst in a tertiary health center in Southwest Nigeria. Pan Afr Med J. 2019 Nov 20;34:154.
- 28. Geller KA, Cohen D, Koempel JA. Thyroglossal duct cyst and sinuses: a 20year Los Angeles experience and lessons learned. Int J Pediatr Otorhinolaryngol 2014;78:264–7
- 29. Ross J, Manteghi A, Rethy K, Ding J, Chennupati SK. Thyroglossal duct cyst surgery: A ten-year single institution experience. Int J Pediatr Otorhinolaryngol 2017;101:132–6.

How to cite this article: erzoug, M. D., Boumelik, M. A., Merad, Y., Matmour, D., Merad, Z., Tahraoui, A., & Megherbi, O. (2023). Early Postoperative Death in a child Undergoing Thyroglossal dust cysts Surgery. Journal of Current Medical Research and Opinion, 6(10), 1766-1769. https://doi.org/10.52845/CMRO/2023/6-10-3