

Thymopharyngeal duct cyst: A case report of seven-year-old patient

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In the pediatric population, neck masses may have congenital or acquired origin and may pose difficulties in diagnosis and management.^[1] Among congenital lesions, branchial cleft cysts, thyroglossal duct cysts, and cysts of thymic origin (thymus cysts and thymopharyngeal duct cysts) stand out.^[2] Embryologic origin, localization, and histopathologic findings are critical in the differential diagnosis of these lesions.^[3] Thymus cysts originate from the remnants of the embryologic thymopharyngeal duct and are usually localized in the anterior mediastinum or lateral neck.^[4] Thymopharyngeal duct cyst is one of the rare benign cervical lesions in the pediatric population. Rare thymopharyngeal duct cysts are associated with fusion anomalies of the third and fourth branchial arches and present as midline or lateral neck masses extending posterior to the pharynx.^[5] In the literature, 10 cases have been published as thymopharyngeal duct cysts.^[6] It is generally accepted as a congenital anomaly and usually proceeds

Abstract

A thymopharyngeal duct cyst is a rare benign cervical lesion in children that arises from remnants of the thymopharyngeal duct that connects the thymus and pharynx during embryonic development. These cysts are typically congenital and asymptomatic, and they are rarely diagnosed unless complications arise. Herein, we reported the case of a seven-year-old male patient who presented with a large, painless mass in the left cervical region that developed slowly over the last three months. The mass became more prominent after an upper respiratory tract infection and was followed by an episode of fainting. On clinical examination, the mass was soft, nontender, and located posteromedial to the left sternocleidomastoid muscle with no signs of inflammation. It was observed to swell during swallowing and coughing. Magnetic resonance imaging revealed a 120x55 mm multilocular cystic lesion extending from the cervical region into the mediastinum and located near the bifurcation of the carotid artery and internal jugular vein. Initially, the mass was suspected to be a hemorrhagic cystic lymphangioma, and bleomycin injection was performed after ultrasonography-guided fluid drainage of 50 mL. However, no significant reduction in size was observed on follow-up imaging. Based on the location and characteristics of the lesion, a diagnosis of thymopharyngeal duct cyst was made. Surgical resection was performed under general anesthesia, and histological examination confirmed the diagnosis. This case represents one of the largest thymopharyngeal duct cysts extending into the mediastinum, and there were no postoperative complications.

Keywords: Cervical lesions, mediastinum, thymopharyngeal duct cyst.

Received: December 21, 2024

Accepted: April 24, 2025

Published online: August 07, 2025

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Citation:

Abilov A, Allahverdieva T, Kurugoglu S, Yusifli Z, Topuzlu Tekant G. Thymopharyngeal duct cyst: A case report of 7-year-old patient. Turkish J Ped Surg 2025;39(3):180-184.
doi: 10.62114/JTAPS.2025.114.

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asymptomatic. However, in some cases, the size of the cyst may increase with infection or trauma and may lead to various symptoms. Herein, we presented a patient with a large, rapidly growing mass in the left cervical region.

CASE REPORT

A seven-year-old male patient presented with a large swelling in the left cervical region that had

suddenly developed over the last three months. The child had no previous health issues. However, it was learned that the mass enlarged after a recent upper respiratory tract infection, after which the patient had a sudden fainting attack. Clinical examination revealed a nontender soft mass located anterior to the left sternocleidomastoid muscle without signs of inflammation. The mass was observed to bulge during swallowing and coughing. Written

informed consent was obtained from the parent of the patient.

Ultrasonography and magnetic resonance imaging results showed a multilocular cystic lesion measuring 120×55 mm on coronal sections at its widest part (Figure 1). The lesion was located medial to the left sternocleidomastoid muscle, close to the bifurcation of the carotid artery and the internal jugular vein, and extended into the mediastinum.

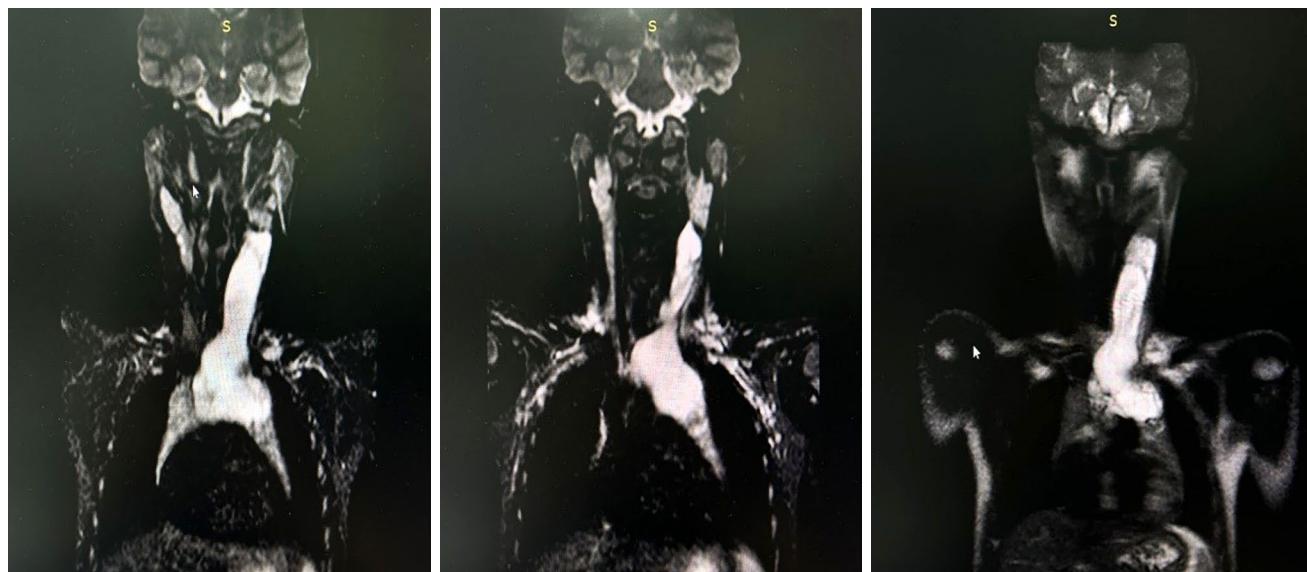


Figure 1. Contrast-enhanced neck MRI examination: A cystic lesion was observed on the left side of the neck, starting between the sternocleidomastoid muscle and the deep cervical tissues and extending inferiorly along the posterior margin. MRI: Magnetic resonance imaging.

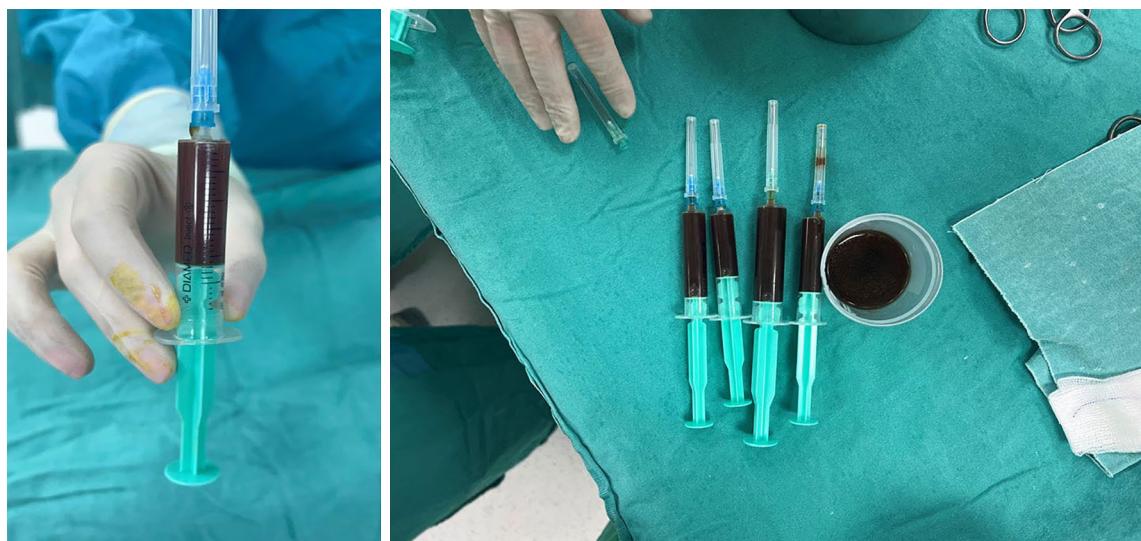


Figure 2. Fluid aspirated from the cyst at the time of bleomycin injection.

The portion of the mass extending into the mediastinum constituted 40% of the total length of the mass. Initially, the mass was evaluated as a hemorrhagic cystic lymphangioma, and bleomycin injection was decided. Under ultrasonography guidance, 50 mL of the fluid in the cyst was drained (Figure 2), and 15 mg bleomycin was injected instead of the extracted fluid by rinsing with 20 mL of 0.9% sodium chloride. However, no significant change in the size of the mass was observed in control examinations. Therefore, considering the anatomical location and pathologic findings, the lesion was thought to be a thymopharyngeal duct cyst. A literature search revealed similar

magnetic resonance images in thymopharyngeal duct cysts.^[7,8] Surgical operation was decided, and the cardiosurgery team was kept ready since the mass extended to the mediastinum.

Under general anesthesia, a left cervical sagittal incision was made over the mass. The cystic lesion was located posterior to the sternocleidomastoid muscle. The lesion was dissected from the internal jugular vein, carotid artery, vagus nerve, and thyroid gland. The part of the mass extending to the mediastinum was excised with traction. It was observed that the mass extended toward the thymus. The mass was completely excised with partial

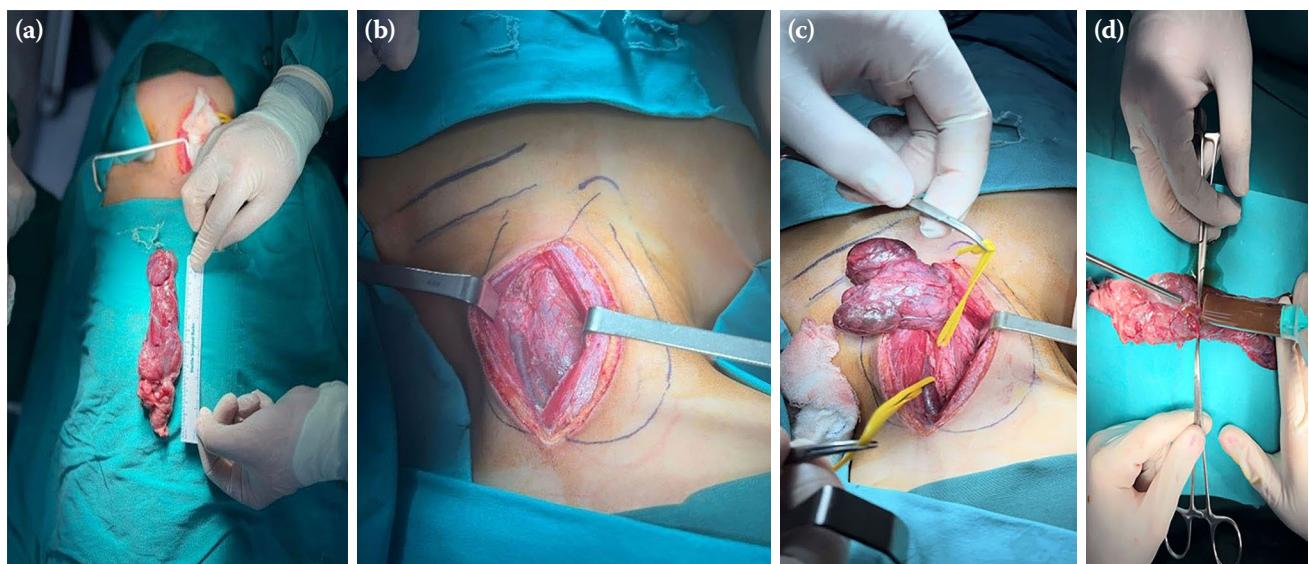


Figure 3. (a) Enlarged neck mass. (b) Cyst; Jugular notch. (c) Cyst; Vagus nerve; Internal jugular vein. (d) Cystic fluid.

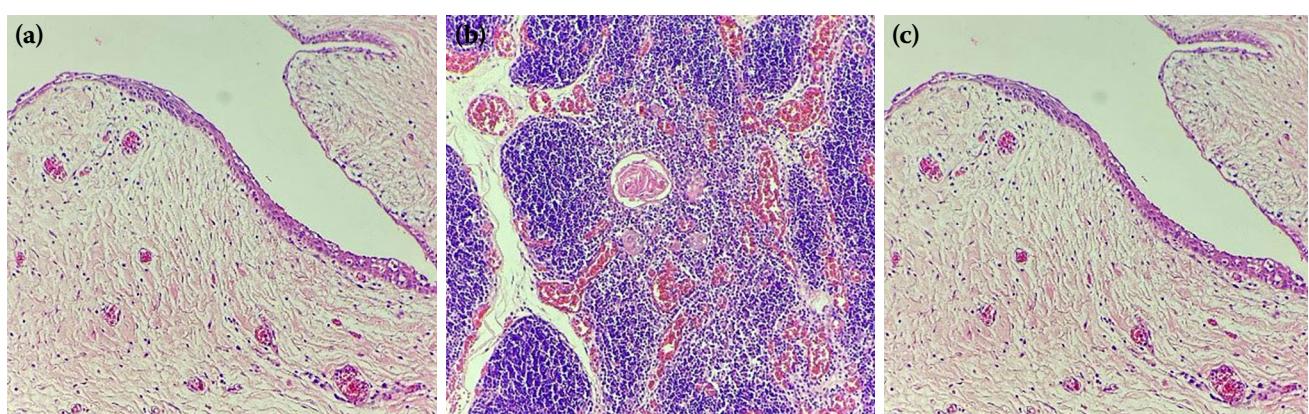


Figure 4. (a) Cyst wall lined by squamous epithelium (H&E, $\times 200$). (b) Thymic tissue showing a Hassall's corpuscle. (H&E, $\times 200$). (c) Thymic tissue within the cyst wall (H&E, $\times 100$).

thymectomy. The total length of the excised surgical specimen was measured as 14 cm (Figures 3a-c). A Minivac drain was placed, and the operation was terminated. There were no complications in the postoperative period.

On macroscopic examination, the maximum diameter of the cyst was 4 cm. Histologic analysis revealed the presence of Hassall bodies, cholesterol granuloma formation and lymphocytic infiltration. The epithelium of the cyst was mostly cuboidal and partially squamous (Figure 4a-c). Immature thymic tissue was not detected. After histologic examination, the diagnosis was congenital thymopharyngeal cyst.

DISCUSSION

Timopharyngeal duct cyst is a rare entity in pediatric surgery. In the literature, it has been reported that these cysts are mostly accepted as congenital anomalies and often have an asymptomatic course. In the study by Smith et al.,^[9] it was reported that thymopharyngeal duct cysts frequently became apparent after infection and trauma. In this case, the enlargement of the cyst and sudden fainting attacks developed during the infection period and made surgery mandatory.^[10]

In a case report published by Johnson et al.,^[11] it was reported that a large thymopharyngeal duct cyst was located in the left cervical region and caused difficulty in swallowing. In this case, it was stated that the size of the cyst reached up to 9 cm, but it was emphasized that it did not extend to the mediastinum. In our case, the size of the cyst reached up to 14 cm and extended to the mediastinum, which is remarkable when compared to other cases in the literature.

In the literature, it is seen that thymopharyngeal duct cyst is frequently confused with hemorrhagic lymphangioma. In the study of Brown et al.,^[12] hemorrhagic lymphangioma was diagnosed in a case with similar clinical findings, but it was found to be a thymopharyngeal cyst with subsequent surgical intervention. In our case, the cyst, which was initially evaluated as a hemorrhagic lymphangioma, was reevaluated as a thymopharyngeal duct cyst because its dimensions did not change after Bleomycin treatment.

Some studies have reported that sternotomy may be required during surgery of large cervical lesions. For example, Boyd et al.^[13] in their article titled "Persistent thymopharyngeal duct cyst" stated that sternotomy may be required for large-sized thymopharyngeal duct cysts. However, in our case, despite the mass extending to the mediastinum, surgical excision could be performed without the need for sternotomy with pressure on the sternum and traction applied to the mass.

In conclusion, this case illustrates the difficulties encountered in the diagnosis and treatment of thymopharyngeal duct cyst and the points to be considered. As a congenital lesion, thymopharyngeal duct cysts require early diagnosis and intervention and exhibit similar findings and results when compared with other cases in the literature^[14] Although rare in neck masses, thymopharyngeal duct cyst is one of the diagnoses that should be considered.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Performed the surgery and contributed to the study concept and manuscript design: A.A.; Assisted in surgery, contributed to manuscript drafting and data collection: T.A.; Radiological diagnosis, image interpretation, and figure preparation: S.K.; Pathology diagnosis, histopathological data interpretation: Z.Y.; Final revision, overall supervision, and manuscript approval: G.T.T.

Conflict of Interest: The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding: The authors received no financial support for the research and/or authorship of this article.

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