

# Large thyroglossal duct cyst of the neck mimicking cervical cystic lymphangioma in a neonate: a case report

Journal of International Medical Research

49(4) 1–8

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DOI: 10.1177/0300060521999765

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## Abstract

Thyroglossal duct cyst (TGDC) is a congenital neck malformation, with a rate of approximately 7% in paediatric patients. TGDC is rarely detected in infants aged younger than 1 year. Even though TGDC is histologically benign, it is associated with preterm delivery or sudden infant death due to airway obstruction, with a mortality rate of 30% to 40%. We report a rare case of a neonate who presented with a large left lateral neck mass. At 7 to 8 months of gestation, magnetic resonance imaging of the foetal neck showed that there was a high possibility of a cervical cystic lymphangioma. The patient had normal vital signs and was afebrile. She was immediately transferred to our Ear, Nose, and Throat Department for further diagnosis and treatment. A computed tomography scan confirmed a large cystic mass that was positioned against a thyroglossal duct. Excision of the mass in the left neck was performed under general anaesthesia without resecting part of the hyoid bone. A histopathological examination confirmed the diagnosis of a TGDC. Follow-up at 1 year showed no recurrence.

## Keywords

Neck mass, thyroglossal duct cyst, neonate, cervical cystic lymphangioma, hyoid bone, inflammation, anaemia

Date received: 28 January 2021; accepted: 8 February 2021

## Introduction

Thyroglossal duct cyst (TGDC) is the most frequently encountered type of congenital anomaly of the neck. TGDCs are formed

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by mucus production in an incompletely degenerated thyroglossal duct. TGDC is mainly only observed if there is secondary fistulation or infection.<sup>1</sup> The gold standard treatment for TGDC is Sistrunk's procedure because of its ability to reduce the possibility of recurrence compared with other surgical treatments and its ability to reveal cystic features in several cases.<sup>2</sup>

Recognition, dissection, and total excision of a TGDC and its surrounding tracts can be carried out without hyoid bone resection, with no recurrence or minor complications. The effect of preserving the hyoid bone on the change in sequence of swallowing and the retroglossal space still needs to be investigated.<sup>3</sup> Additionally, approaching a full thyroglossal duct (TGD) via the classical operation in children is difficult. This is difficult because finding the duct, which appears soft, is impossible owing to the location of the TGD at the tongue base and its remote location from the transcutaneous incision site. Taking into account the external surgery scar and embryological features, other surgical methods need to be introduced that provide access to the complete TGD and leave no scar.<sup>4</sup>

Power and colour Doppler applications have enabled good diagnostic utility for TGDC. The extent of the lesion, size, fat, and calcification of the lesion, and its association with surrounding structures is ruled out by ultrasound and computed tomography (CT). Magnetic resonance imaging (MRI) also has an extra multi-planar diagnostic capability for more chronic and locally broad lesions. Fine-needle aspiration cytology can be carried out to confirm a diagnosis in a few cases. Because the risk of infection with TGDC is high, under elective conditions, cysts should be removed. The factors that raise the risk of recurrence are insufficient treatment, infected cysts, incorrect diagnosis, drainage of the cyst, perforation of the cyst during treatment, and patients aged younger than 2 years.<sup>5,6</sup>

After prenatal diagnosis, follow-up should be performed to determine growth, characteristics, and the size of the cyst.

We report a rare case of a neonate who presented with a large TGDC. Excision of the mass in the left neck was performed and follow-up at 1 year showed no recurrence.

## Case report

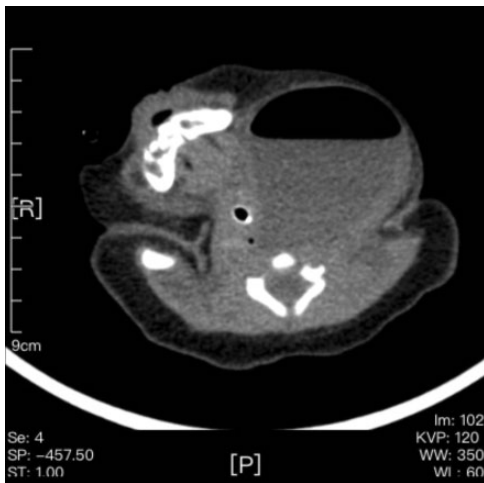
A female neonate was admitted to our department because of a mass in the left lateral neck (Figure 1). She is the only child of her mother and was born at 39<sup>1/7</sup> weeks of pregnancy. She was delivered by selective caesarean section with a birth weight of 3.59 kg. At 7 to 8 months of gestation, foetal neck MRI showed that there was a high possibility of a cystic cervical lymphangioma. Before the diagnosis, she had no history of intrauterine distress or postnatal asphyxia, and amniotic fluid, the umbilical cord, and placenta were normal. She had an Apgar score of 10 at both 1 and 5 minutes after birth. She was transferred to our department for further diagnosis and treatment. A physical examination on admission showed that her general state was good, her skin was ruddy, and the anterior fontanelle was flat without tension. A left cervical mass of 5.0 × 4.0 × 4.0 cm was found with a clear



**Figure 1.** Preoperative view of the patient.

boundary, it was soft on palpation, and there was a normal skin colour on the surface. The patient's respiratory rate was normal, there were no abnormal breath sounds, and dry and wet rales were not heard. Her heart rate was 140 beats per minute and the rhythm was regular with no heart murmurs. During the hospital stay, the child frequently required mechanical ventilation at night. Neck MRI could not be completed because of poor cooperation of the child. The mass was gradually increasing with airway compression.

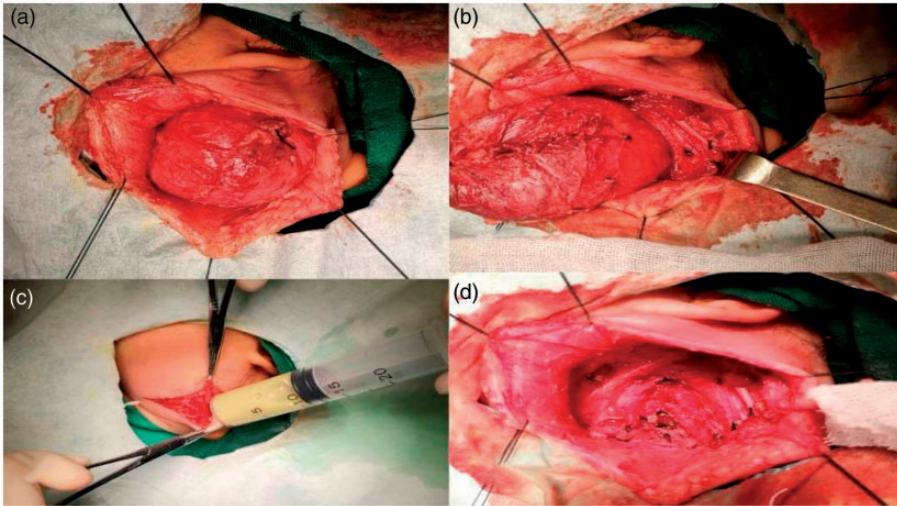
Ultrasound (US) of the left neck mass showed a thick-walled cystic echo, which was observed on the left side, and the size of the mass was  $56.6 \times 32.9 \times 33.2$  mm with poor internal transparency. The deep part of the mass was located beside the trachea behind the left lobe of the thyroid gland, and dense punctate hyperechoic foci were observed on the back wall. The shape of the mass changed with the body position. A CT scan of the left cervical neck showed a large cystic mass that was positioned against a thyroglossal duct (Figure 2). The mass had compressed the



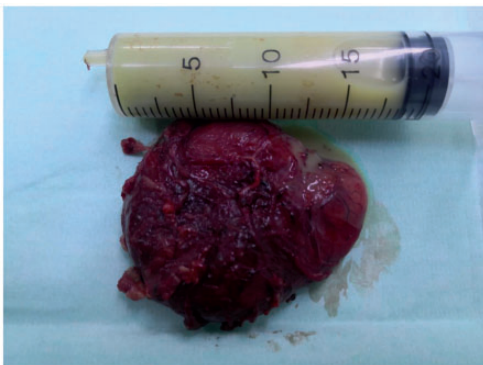
**Figure 2.** Computed tomography image shows an extremely large cystic lesion, extending laterally towards the left.

trachea and pharyngeal cavity, and obstructed breathing. Because of the neonate's extremely large mass, endoscopy/laryngeal examination was not carried out. Surgical resection of the neck mass was performed with a transverse incision of 6 cm. A volume of 17 mL of sac fluid was aspirated with a 20-mL syringe to reduce the mass size (Figure 3a–d). A few soft tissues on the surface of the hyoid bone were resected with care to prevent injury to the nearby neurovascular bundles of the neck, trachea, and oesophagus. The cystic mass, which was  $5 \times 4.5 \times 2.5$  cm in volume, 5 cm in diameter, 0.1 to 0.3 cm in wall thickness, and light yellow with turbid liquid, was successfully excised (Figure 4). A histopathological examination confirmed the diagnosis of a TGDC with suppurative inflammation. Most of the cyst wall had epithelial necrosis and a large amount of necrotic and inflammatory exudation in the cavity. Acute and chronic inflammatory cell infiltration with a small amount of thymic tissue were observed around the cyst wall (Figure 5a and b). The patient's diagnosis was a TGDC with suppurative inflammation.

Surgical removal of the neck mass that originally compressed the trachea posed a risk of the trachea to soften. Therefore, mechanical ventilation was continued. On the 10th day after surgery, the patient had a fever, an increased leukocyte count, and the high-sensitivity C-reactive protein level was 16.33 mg/L. These findings suggested the presence of an infection, and therefore, meropenem was administered. Sputum culture was performed and it showed positivity for *Staphylococcus aureus* and *Enterobacter cloacae*. *E. cloacae* is sensitive to cefepime. Therefore, vancomycin was added for anti-infection treatment. The neck incision appeared to have exudation. We considered performing superficial tissue US to confirm this possibility. For the postoperative



**Figure 3.** Surgical resection of the neck mass was performed with a transverse incision of 6 cm. A volume of 17 mL of sac fluid was aspirated with a 20-mL syringe to reduce the size of the mass.



**Figure 4.** An excised cystic mass of  $5 \times 4.5 \times 2.5$  cm in volume, with a diameter of 5 cm, and wall thickness of 0.1 to 0.3 cm. The lesion is slightly yellow with turbid liquid.

exudation, the dressing was routinely changed and it then gradually decreased.

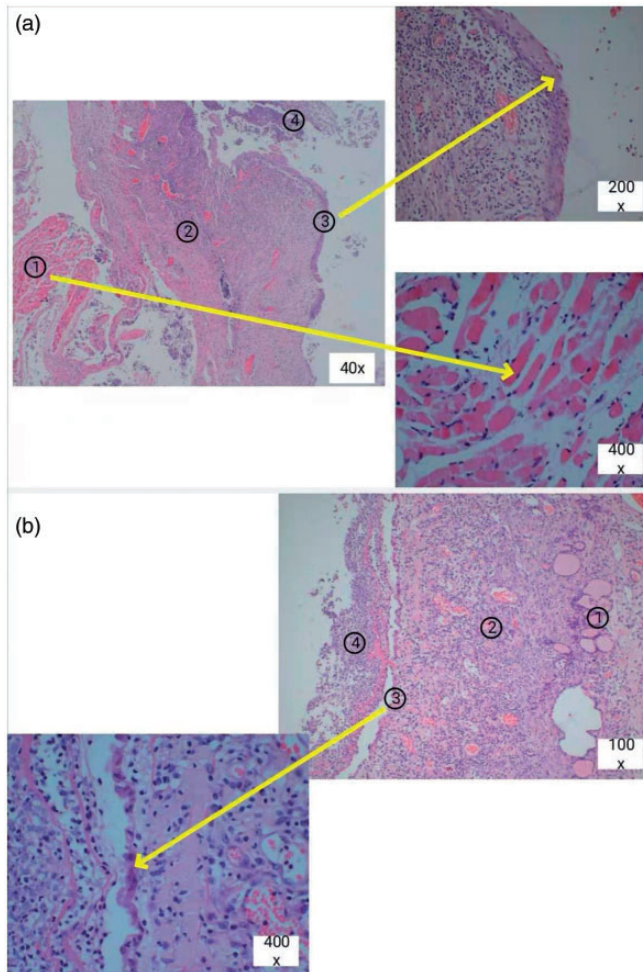
Routine blood tests showed that haemoglobin levels were  $<145$  g/L. Therefore, the supplementary clinical diagnosis was neonatal anaemia. The degree of anaemia was not serious and no blood transfusion was required. On the 11th day after the operation, a transverse incision of 6 cm was

visible in the left neck without exudation. Respiratory dyspnoea was observed; therefore, bronchoscopy was performed, which confirmed malacia of the retropharyngeal cavity. This indicated a high possibility of respiratory infection and pneumonia. Long-term compression of the cyst might have caused local peripheral tissue dysplasia. Therefore, symptomatic treatment was added, including calcium, phosphorus, and alkaline phosphatase. The patient was successfully discharged with oral medication of vitamin AD capsules (500 IU) and vitamin D1 capsules. Alternatively, she could take oral vitamin AD, calcium zinc gluconate, bicyclol 5 mg, and *Clostridium butyricum* in case of poor defecation. At 1 year of follow-up, the patient was well with no evidence of recurrence.

## Discussion

Cystic masses in the neck have a number of pathological entities. The age of presentation and clinical assessment can be used for differential diagnosis, but imaging is necessary for accurate diagnosis and





**Figure 5.** Pathological results confirming a thyroglossal duct cyst. (a) (1) Striated muscle around the cyst wall; (2) inflammation in the cyst wall and inside the cyst wall; (3) the remnant lining of the cyst wall is lined with stratified epithelium (non-vascular endothelial morphology); and (4) inflammatory exudation in the cyst cavity. (b) Because of various factors, such as a limited field of view, extremely large cyst volume, and destruction of the epithelium and thyroid tissue in the cyst wall by inflammation, we were unable to view the lining epithelium. The thyroid tissue in the cyst wall and striated muscle around the cyst wall were continuously and completely expressed in one field of vision.

pretreatment preparation.<sup>7</sup> Nevertheless, TGDC is a rare condition in the paediatric age range.<sup>8</sup> Diagnostic imaging with US followed by CT and MRI examinations help in diagnosis and evaluation of the anatomical area and complications, including pre-treatment planning. MRI or CT is favoured because it can show an orthotopic thyroid

gland and be used to assess and distinguish features and a range of neoplastic mechanisms. A neck cyst histologically comprises pseudostratified ciliated columnar epithelium and may or may not involve ectopic thyroid gland tissue.

TGDC is a rare occurrence and has an incidence rate of only 1%.<sup>9</sup> If TGDC occurs

*de novo* from native thyroid tissue in the cyst wall or as a metastasis from the thyroid gland, the origin of TGDC is unclear.<sup>9</sup> CT, US, and MRI features can often be equivocal for diagnosing TGDC. Laryngocele, dermoid/epidermoid cyst, branchial cleft cyst, squamous cell carcinoma, lateral branchial cyst, pyriform sinus fistula (PSF), abscess, thymic cyst, lymphatic malformation, and metastatic disease are some of the differential diagnoses. Even though a close relationship of TGDC with the hyoid bone is an important characteristic of differentiation, conclusive differentiation is mostly not achieved until pathological diagnosis.<sup>10</sup> Only a few studies have described PSF that was diagnosed neonatally or prenatally.<sup>11</sup> In these fetuses, a cystic mass in the left side of the neck and presentation with a cervical cystic lesion due to the effect of compressed infection and symptoms were observed.<sup>11</sup> Approximately 80% of PSF appears in older children and its incidence is the same in both sexes. PSF begins at the pyriform sinus apex of the hypopharynx, and it invades the cricothyroid muscle and ends in or adjacent to the dorsolateral part of the left lobe of the thyroid.<sup>12</sup> However, TGDC arises from the embryonic remnants because of inability of the TGD to close at the foramen cecum to the area of the thyroid in the neck.<sup>10</sup> Many patients appear to have a history of recurrent neck inflammation, which indicates a high possibility of a PSF.<sup>13</sup>

Clinical characteristics of TGDCs include symptoms of dysphagia, upper airway obstruction, swelling that was located since birth in the midline of the neck, with redness surrounding the swelling, and sporadic discharge from this swelling.<sup>14</sup> During development, the TGD loops inferiorly over the hyoid bone. As a result, a cystic lesion closely related to the hyoid bone can suggest diagnosis of TGDC.<sup>15</sup> Therefore, recognising the embryological path of thyroid formation is important to help differentiate TGDC from other neck

masses with knowledge of essential landmarks, such as the hyoid bone, strap musculature, the foramen cecum, and thyroid cartilage.<sup>16,17</sup> A TGDC can occasionally have a rare presentation either radiologically or clinically, which can be a diagnostic challenge. Not expecting the possibility of a TGDC can be related to performance of a deficient surgical operation, such as enucleation or a simple incisional biopsy, both of which can cause high recurrence rates.<sup>18</sup> Most otolaryngologists have insufficient knowledge of this disease. TGDC should be considered as one of the important differential diagnoses of a neck mass in children.

In our case, we did not resect part of the hyoid bone because the cyst was large, and it was severely adhered to the surrounding tissues and the surface of the hyoid bone. Therefore, finding a relationship between the cyst and the hyoid bone can be difficult.<sup>19</sup> The appearance of a cyst during the operation also supported the preoperative diagnosis. Therefore, we did not resect the affected part of the hyoid bone. Fortunately, the patient had no relapse. In case of relapse, resection of part of the hyoid bone would be required.

The presence of a neck lump before diagnosis does not indicate malignancy. Our patient had her neck mass found before birth. In our case, the situation was interesting because the patient's symptoms appeared during infancy. Nonetheless, rapid growth, a hard mass, and uneven or new changes in the mass may indicate malignancy.<sup>2</sup>

TGDCs are usually typically 2 to 4 cm. Larger cysts are not common and are usually found in adult patients, possibly owing to the gradual continuous growth of these lesions. TGDCs are mostly observed as cysts. Although sporadic and genetic variables play a major role in development of TGDCs, they are considered to sporadically evolve. Even though TGDCs are of embryonic origin, rare symptomatic cases have

been reported during infancy.<sup>20</sup> In neonates with minimal clinical doubt for malignancy, imaging assessment can start with US. However, there might be abnormal sonographic features, such as atypical vascularity, a hard component, or high clinical doubt for malignancy.<sup>21</sup> In this case, Peretz *et al.*<sup>2</sup> advised that the guidelines of TGDC in neonates need to be aggressive, despite the degree of this disease and engagement of lymph nodes. A detailed case history is important in the differential diagnosis.<sup>22</sup> The appearance of internal debris, multiple punctate hyperechoic foci, and septation are commonly associated with early infection in TGDC.

## Conclusion

The most prevalent congenital masses found in the neck are TGDCs. TGDC occurs often in the first decade of life. Although TGDC is often found at an average age of 5 years, uncommon cases can also be observed with symptoms in infancy, as in our patient. A detailed assessment of lateral neck masses requires knowledge of the embryological path of thyroid growth for awareness of TGD anomalies, modifications, and complications. The ability to differentiate characteristics of other cystic neck masses is important because of various clinical implications.

## Author contributions

NF, LNA, YC, and XW searched the relevant literature and helped treat the patient. The final work was approved and read by all authors.

## Declaration of conflicting interest

The authors declare that there is no conflict of interest.


## Ethics statement


Ethics committee approval was not required. The patient's guardian provided written informed consent for publication.


## Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

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