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CASE REPORT

Thoracoscopic management of giant thymolipoma

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Abstract
Thymolipomas are rare benign thymic neoplasms that contribute 2 to 9% of thymic tumors. Surgical excision remains the primary treatment, with video-assisted thoracoscopic surgery (VATS) emerging as a valuable approach. Less than 50 cases of pediatric thymolipomas have been reported, with the youngest case documented at 6 months of age, and fewer than five pediatric cases have been managed using the thoracoscopy approach in the literature. This is India's first report in literature where giant thymolipomas have been excised by the thoracoscopic approach in the pediatric population. Herein, we present two pediatric patients with anterior mediastinal masses, each presenting with distinct clinical features undergoing successful thoracoscopic resections. In case 1, a 9-year-old male child was incidentally diagnosed with giant thymolipoma. Surgical excision was done through the right side by VATS approach and the residual mass through the left by VATS, resulting in an uneventful postoperative course with no recurrence upon 1-year follow-up. In case 2, a 5-year-old female previously diagnosed with giant thymolipoma presented with fatigability. Imaging revealed a massive thymolipoma occupying both hemithoraces. Careful dissection, preservation of vital structures, and successful vascular pedicle management ensured complete excision through a single left thoracic approach, leading to a smooth recovery and absence of mediastinal mass on follow-up X-ray. This case report highlights VATS as a safe and effective management for giant thymolipoma; though it is technically challenging, it is a feasible approach, contributing to improved patient outcomes.

Keywords Pediatric thymolipoma · VATS thymectomy · Minimally invasive surgery · Thymic tumors · Myasthenia gravis · Giant thymolipoma

Introduction

Thymolipoma is an exceedingly rare and distinctive type of benign neoplasm that originates in the thymus gland, constituting approximately 2 to 9% of all thymic neoplasms [1]. It is a subtype of benign anterior mediastinal tumors characterized by a unique amalgamation of thymic and adipose tissues [2]. The incidence of thymolipoma is around 0.12 out of 100,000 cases per year, with a higher incidence in the younger age population [1]. Most of the reported cases showed an association with myasthenia gravis; the prevalence of myasthenia gravis associated with thymolipoma is 43.8% [3].

The definitive treatment for thymolipomas is surgical, with various approaches such as thoracotomy, video-assisted thoracoscopy, and sternotomy, and the decision depends on the size and location of the tumour [4].

Video-assisted thoracoscopic surgery (VATS) is an emerging approach in the treatment of thymolipoma; it is safe, effective, less painful, cosmetic, and allows early discharge of the patient, and no cases have been reported on recurrence. According to Otido et al.'s study, only two studies of thoracoscopic resection of thymolipoma in pediatric thymolipoma cases have been reported in the literature [5]. In the Indian context, no cases have been reported employing thoracoscopic techniques for giant thymolipomas, which highlights the limited exposure to this surgical modality in the region.

In this case series, we present two cases of young pediatric patients diagnosed with giant thymolipomas, effectively managed through the VATS approach. This is India's first report in literature where giant thymolipomas have been excised by a thoracoscopic approach. The success of

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thoracoscopic excision in our cases underscores its feasibility, efficacy, and safety, providing a promising alternative in the surgical management of thymolipomas, with a better prognosis. They not only highlight the clinical features associated with giant thymolipomas, but also underscore the critical role of VATS surgical intervention in achieving favorable outcomes.

Case reports

Case 1

A 9-year-old male child was referred to our medical facility due to a constellation of clinical symptoms, which included lower respiratory tract infection (LRI) and ptosis of the left eye. There was no history of associated fever, haemoptysis, cyanosis, weight loss, dysphagia, or abnormal body movements. Furthermore, there was no prior history of hospitalization, allergies, or asthma attacks.

Initial assessment revealed the presence of an anterior mediastinal mass, which was further complicated by the occurrence of ptosis and mild respiratory symptoms. In an effort to determine the underlying etiology, myasthenia gravis was initially suspected. However, diagnostic tests, including the fatigability test, ice pack test, repetitive nerve stimulation test, and anti-choline esterase antibody levels, returned negative results, effectively ruling out myasthenia gravis as the diagnosis. Subsequently, a comprehensive assessment led to the diagnosis of congenital ptosis, which accounted for the ptosis of the left eye.

Chest computerized tomography (CT) (Fig. 1) of the thorax revealed a large mass measuring 22.5 cm × 15.4 cm × 11.5 cm fat attenuated (−80 to −120 HU) with minimally enhancing internal densities noted in the anterior mediastinum extending downwards onto the left and right *mediastinal* regions, partially wrapping around the heart and obscuring both the borders of the heart. The mass was predominantly of fat density with multiple internal non-homogenous areas of soft-tissue density with no definite pattern. The mass was draped around the heart and great vessels.

Considering the extensivity of the mediastinal mass and the probability of compressive symptoms in the near future owing to the propensity of progressive increase in the size of the lesion, the child was subjected to VATS for the giant thymolipoma excision. The right thoracoscopic approach was chosen. The surgical field was prepared with 5-mm ports, a 30-degree scope, CO_2 insufflation at 6–8 mm Hg, and tissue dissection utilizing a bipolar energy device. The camera port was positioned at the 4th intercostal space in the anterior axillary line, and the working ports at the 3rd and 6th intercostal spaces in the mid and posterior axillary lines, respectively. The dissection commenced at the inferior horn along the phrenic nerve, and the huge lesion was methodically lifted off the pericardium and away from the sternum, along the superior vena cava (SVC) until its confluence with the innominate vein was reached. The cervical horns were isolated, ligaments divided, and venous branches sealed and cut. Posterior dissection led to the identification of the left phrenic nerve. The mass on the left side was delineated, and vascular pedicles were cauterized and divided. The mass on the left hemithorax was guided to the right mediastinum (Fig. 2a). The inferior port was enlarged to accommodate an endobag for specimen removal and orientation for pathology assessment through the right mediastinum. Re-evaluation revealed a total excision of the right *mediastinal* mass and near total excision of left *mediastinal* mass. The child underwent a left thoracoscopy to remove the remnant left *mediastinal* mass.

Histopathologic evaluation of the excised mass revealed a giant thymolipoma. Follow-up X-ray revealed no mediastinal mass (Fig. 3a). The child is recurrence-free on follow-up for 1 year.

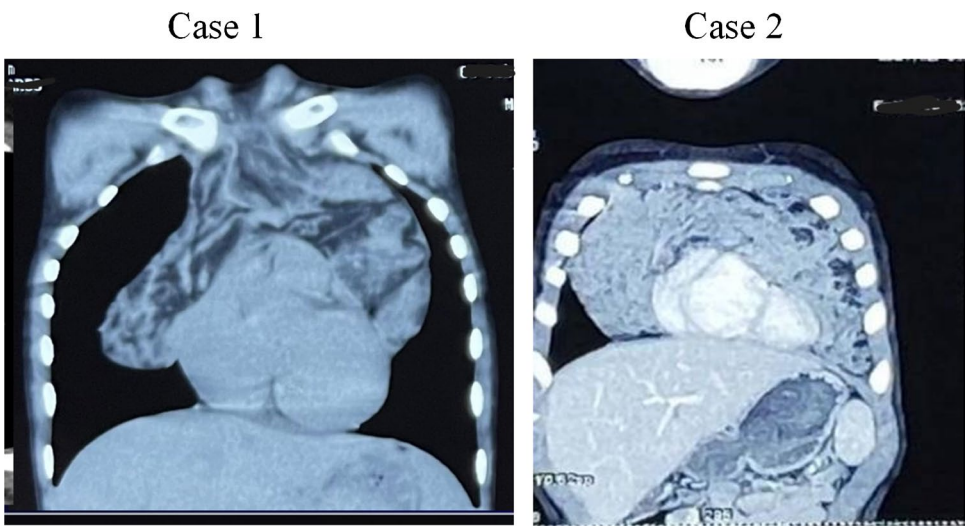


Fig. 1 **(a)** CT image of case 1, (**b**) CT image of case 2

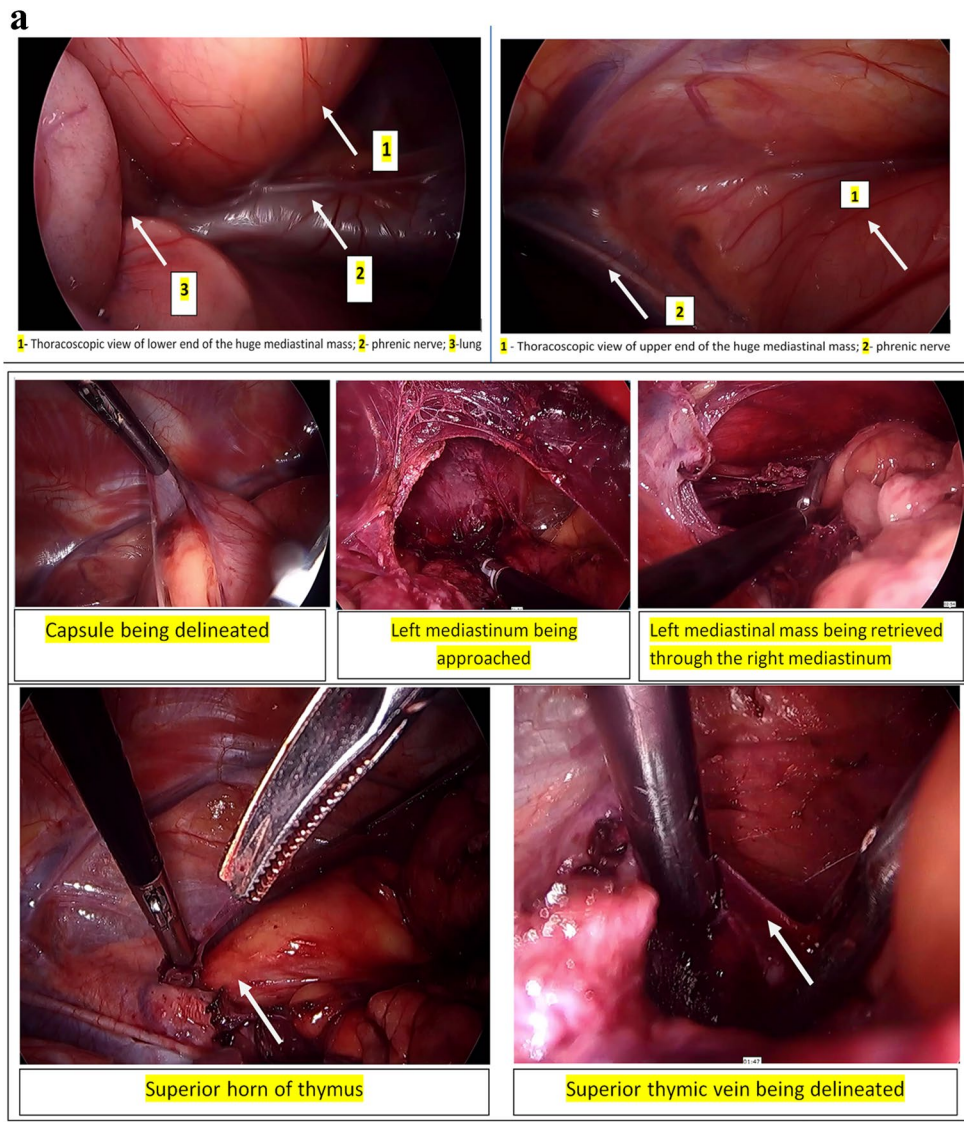
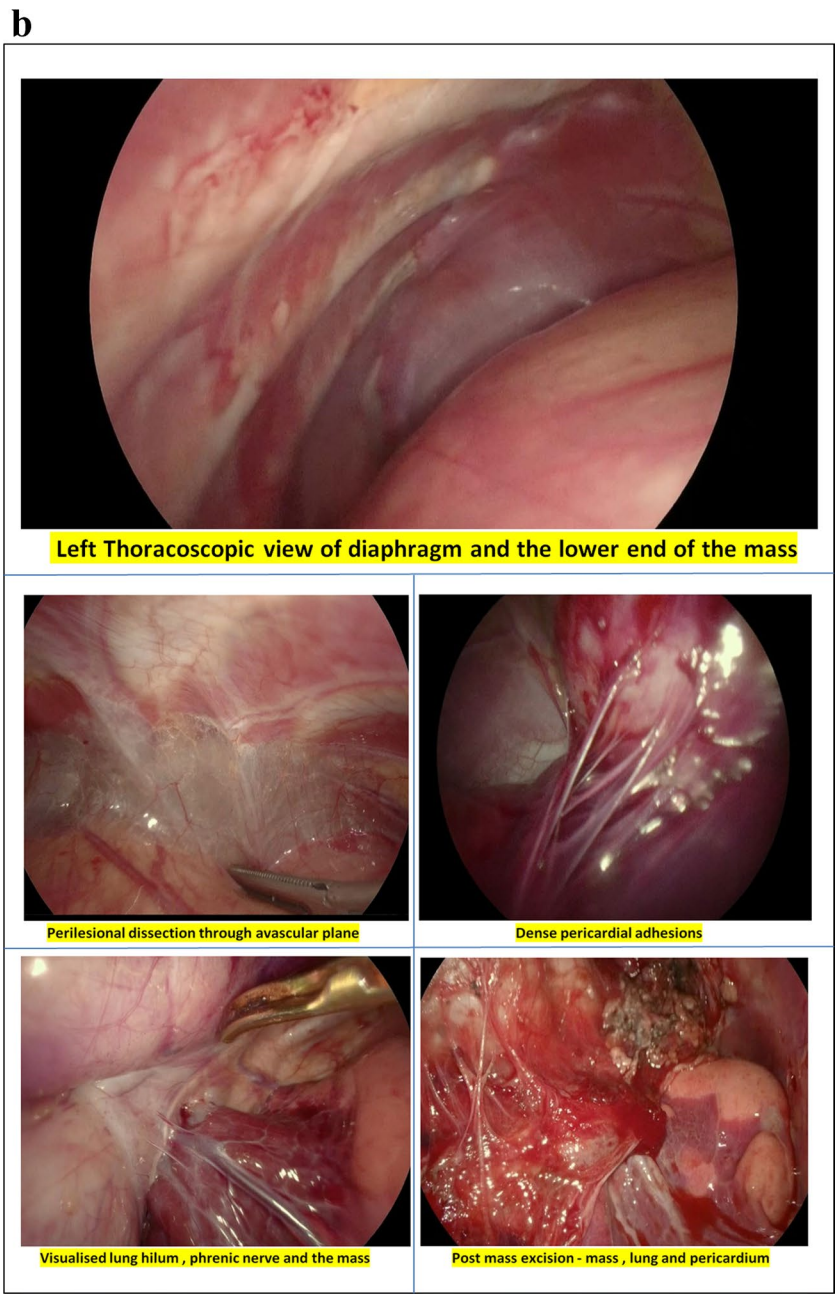


Fig. 2 Computed tomography images of case 1 and case 2. **(a)** Intraoperative images of case 1 and (**b**) Intraoperative images of case 2

Case 2

A 5-year-old female child, previously diagnosed with giant thymolipoma, presented to the hospital with a history of easy fatigability over the past year, particularly after minimal to moderate physical activity. The initial discovery of the condition occurred incidentally during admission to an outside hospital for dengue fever. A chest X-ray (Fig. 3b) revealed complete opacity of the bilateral chest with minimal sparing of the left apex, prompting further investigation through a CT chest (Fig. 1). The patient denied any history of fever, loose stools, ear discharge, rashes, urinary disturbances, altered level of consciousness, seizures, or bleeding manifestations. Upon examination, the child had stable vitals. Respiratory examination indicated reduced air entry on the left side. CT scan of the chest revealed a bilateral mediastinal heterogeneous mass left to right (L-R) lesion with soft tissue density and streaks of fat in the anterior mediastinum. The size of the lesion measured 7.5×16×11 cm (APxTRxCC), raising suspicion of giant thymolipoma or liposarcoma. The patient was admitted to the hospital for the surgical resection of the giant thymolipoma and planned for thoracoscopic excision. The left thoracic cavity was entered through 5-mm ports at the 3rd and 5th intercostal spaces over the anterior axillary vein. During the procedure, a massive thymolipoma was observed, occupying the entire left hemithorax. Dense adhesions were noted between the lesion and the pericardium. With visualization and preservation of the phrenic nerve, the lesion's capsule was delineated. There were also dense adhesions to the left lung hilum, which was delineated. The vascular pedicles were clearly delineated from the subclavian and brachiocephalic vessels, clipped/cauterized and divided. The mediastinal window on the right side was entered through the left thoracoscopic window, and the mass was visualized and delineated (Fig. 2b). The vascular pedicle on the right

Fig. 2 (continued)



was cauterized and divided under vision. The right phrenic nerve was preserved, and the entire mass, weighing 600 g, was excised with no residual lesion. The post-operative period was uneventful, and the child was discharged on POD 4 (post-operative day 4). Follow-up X-ray revealed no mediastinal mass (Fig. 3b).

Discussion

Pediatric thymic lesion, especially thymolipoma, is exceptionally rare [1]. It was first reported by Lange in 1916 and the term "thymolipoma" was first coined by Hall in 1948 [6]. These benign tumours originate from thymic fat, and they are characterized by mature thymic tissue and adipose components [4]. These tumours can enlarge, mould, and accommodate in the thoracic cavity, often achieving massive dimensions. Less than 50 cases of childhood thymolipomas have been reported [7]; among these, the youngest was 6 months old, and it is the only case reported on infants [6]. According to Otido et al.'s study, only two studies of thoracoscopic resection of thymolipoma in pediatric thymolipoma cases have been reported in the literature [5]. There is no discernible gender predilection. Our literature searches encompassed databases such as PubMed and Google Scholar, utilizing MeSH

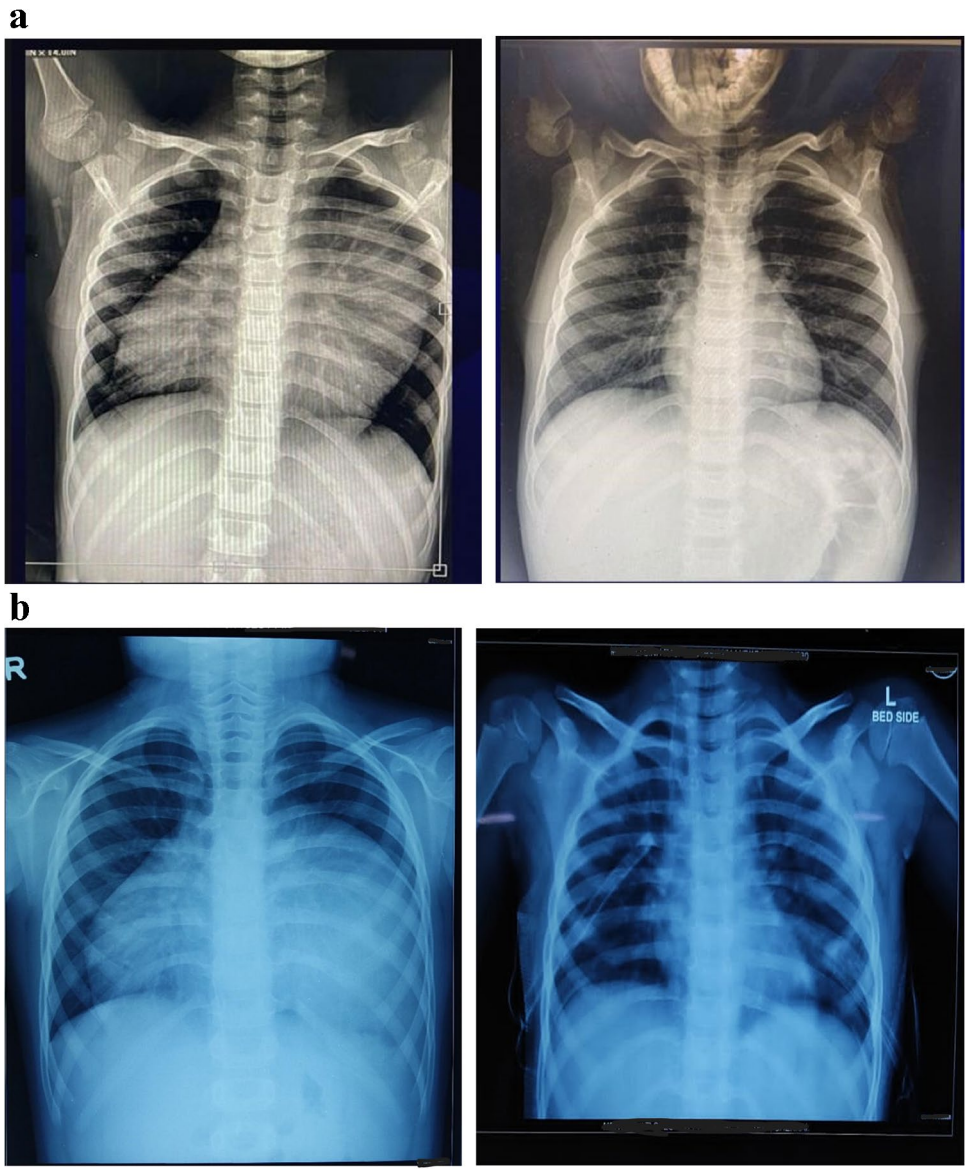


Fig. 3 (**a**) Preoperative and postoperative X-Ray of case 1, (**b**) Preoperative and postoperative XRay of case 2

terms and keywords such as "Thymolipomas," "pediatric thymolipoma," "Thoracoscopy," "VATS in pediatric thymolipoma," "Pediatric thymolipoma," and "Infant Thymolipoma."

The pathogenesis of thymolipomas is still unclear. Recent data suggest a potential link with mutations in the 12q15 high mobility group AT-hook 2 (HMGA2) gene [7]. Several hypotheses have been put forward, such as simple thymic and fatty hyperplasia, actual mixed neoplasms of endocrine and mesodermal origin, or fatty neoplasms that subsequently lead to thymic involution [6].

Most thymolipomas are indolent, and they are found incidentally [7, 8]. Some patients report symptoms of upper respiratory tract infection (RTI), chest discomfort, dyspnea, tachypnea, and non-specific chest symptoms. Some patients have a history of insufficient weight gain. Few studies have indicated its associations with myasthenia gravis, especially in adult patients; other related illnesses include aplastic anaemia, Graves' disease, Hodgkin's disease, Gardner's syndrome, pure red blood cell aplasia, lichen planus, aplastic anemia, and hypogammaglobulinemia [3, 5, 7, 8]. In our cases, the diagnosis of thymolipoma was made incidentally; this underscores the importance of considering thymolipoma as a potential finding, even in the absence of typical clinical syndromes, and highlights the significance of early detection and diagnosis in such scenarios.

Thymolipomas are diagnosed by the presence of a fatty mass, their location, and continuity with the thymus [7]. Diagnosing thymolipoma based on clinical symptoms or signs is challenging, and therefore, the definitive diagnostic method is considered to be a biopsy [3]. It is mostly asymptomatic and often found incidentally by chest radiography;

it is therefore recommended that initial radiological assessment should include neck and chest radiographs. Currently, CT is the preferred imaging modality to evaluate mediastinal masses. It differentiates benign thymic lesions from thymic malignancies [6, 8]. The differential diagnosis includes thymoma, thymohemangiolipoma, lipomas of thymic origin, mature thymic hyperplasia teratoma, and rarely, well-differentiated liposarcomas [5, 7].

In view of its associations with myasthenia gravis, immune disorders, unusual variants, and small risk of malignancy, surgery should be considered in all cases. In our patients, VATS excision was performed. Coosemans et al. first performed this in 1993 [9]. Although the routine mode of approach is open thoracotomy and sternotomy [5], we have adopted thoracoscopy; though challenging, it is more rewarding because of its advantages like reducing surgical trauma, less intraoperative blood loss and duration of postoperative pleural drainage, less postoperative pain, reduced hospital length of stay, better result, rapid recovery, and lower complications.

Numerous surgical options are available, and the final choice depends on the individual surgeon's preferences and expertise. In VATS, the choice of the first side of the thorax to be approached varies according to the surgeon's experience and preference. The intraoperative steps may also vary [10]. Recurrences have not been reported in any of the studies.

Conclusion

This is India's first reported case series in literature where giant thymolipomas have been excised by the thoracoscopic approach in the pediatric population. VATS proves to be safe and effective and offers advantages such as reduced surgical trauma, less intraoperative blood loss, shorter postoperative pleural drainage, postoperative pain, shorter hospital stays, and quicker recovery. While conventional open thymectomy has been the traditional gold standard, VATS reflects a paradigm shift towards minimally invasive techniques, showcasing its adaptability and success, even in giant thymolipomas. Continued exploration of VATS in pediatric giant thymolipoma excisions is essential to establish its role further and encourage broader adoption in clinical practice.

Learning points

- Pediatric thymolipoma, a rare, benign thymic-origin mediastinal mass, can be asymptomatic despite its substantial size; around 50% are linked to autoimmune diseases.
- Diagnosis involves clinical assessment, imaging, histopathological biopsy, alpha-fetoprotein (AFP) level testing, and considering associated autoimmune disorders.
- Differential diagnoses encompass various mediastinal masses, making comprehensive evaluation and biopsy vital for differentiation.
- VATS, though technically challenging, provides excellent prognosis when managed by experts.

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Author contribution RM participated in the data acquisition, surgical planning, design operative execution, literature review, manuscript preparation, and manuscript editing. HG participated in the data acquisition, surgical planning and design, operative execution, and manuscript editing. VD participated in literature search, surgical planning, operative execution, and manuscript editing. RSM and SS participated in investigations, surgical planning, and surgery execution.

All authors have approved and approved the manuscript.

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Declarations

Ethical committee approval Not applicable.

Informed consent Informed consent was obtained from all the patient's parents.

Competing interests The authors declare no competing interests.

Statement of human and animal rights This study upholds the principles of the Declaration of Helsinki for human subjects and follows the Ethical guidelines, ensuring informed consent, confidentiality, and ethical treatment in accordance with established standards.

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