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Incidentally detected giant pediatric mediastinal mass in a young girl primarily treated for dengue

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Abstract
Most of the paediatric mediastinal masses are detected incidentally when chest radiographs are done on asymptomatic children or on those presenting with respiratory symptoms. We hereby report a case of a young girl who was primarily treated for dengue fever and an incidental giant mediastinal mass was detected when chest x ray was done for her respiratory symptoms. Computed tomography (CT) scan of the chest and excision was done which proved it to be a giant benign mesenchymal tumor of the mediastinum. Histopathological diagnosis and surgical excision remains the main stay of treatment.

Keywords: Mediastinum, asymptomatic, benign mesenchymal tumor, giant

Introduction
Thymus which is located in the anterior mediastinum is an important part of immune system in paediatric population. Thymic lesions are rare and account for approximately 2% of all mediastinal tumors [1]. A wide histological spectrum of lesions are seen in mediastinum due to presence of many organs in the mediastinum. Examples include congenital lesions, infections, vascular lesions, benign and malignant lesions [2]. There are diverse causes of anterior mediastinal masses in children of which thymoma, lymphoma and teratoma are the commonest. Here is a case report of a massive benign asymptomatic mesenchymal tumor involving bilateral thorax indenting lung parenchyma and pericardium in a young girl primarily admitted for dengue fever.

Case Report
A 5 year old developmentally normal female child was brought to emergency room with history of high grade intermittent fever, running nose and poor oral intake. The child was earlier treated elsewhere and was found to have dengue IgM positive with thrombocytopenia. Chest X ray was done which showed complete left lung opacification with minimal sparing of apex with no tracheal or mediastinal shift. Contrast enhanced CT was advised which revealed a well defined mediastinal mass with intermingled macroscopic fat, more on left side, maximum thickness measuring 5.5mm. There was no calcification. Tumour markers (alpha feto protein and beta hcg) were negative. Hence planned for excision and histopathological correlation.
The child was taken up for Thoracoscopic excision of the giant lesion. Intra operatively there was a huge mass occupying the entire left hemithorax with dense adhesion to the pericardium and the left lung hilum. Dense peritumoral adhesions were released by a combination of blunt and sharp dissection. The capsule was opened, left phrenic nerve preserved and the mass was excised in toto from the right mediastinum too preserving the right phrenic nerve. Vascular pedicles from the subclavian, brachiocephalic and internal thoracic were cauterised and divided. No residual mass lesion was identified on thoracoscopic visualisation on either hemithoraces. Post operatively child is on follow up for 6 months. The child is asymptomatic and no evidence of recurrence noted.

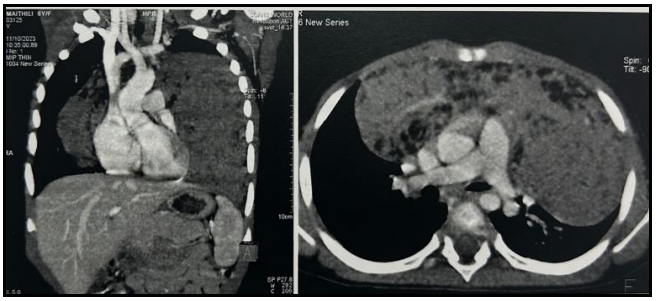

Fig 1: A large well defined lesion with soft tissue density and streaks of fat in the anterior mediastinum. (ultra low dose Multi slice CT - Chest)

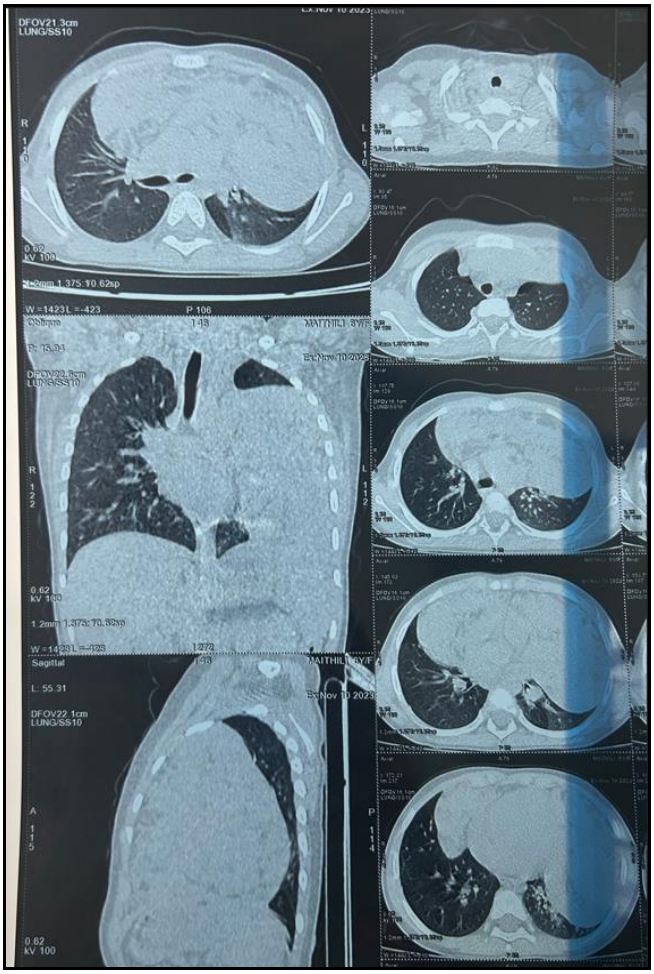

Fig 2: Oblique and saggital view showing a well defined large heterogeneous predominantly soft tissue densityfocus with interspersed fat seen with its epicenter in the mediastinum extending into bilateral thorax (L>>R) indenting the lung parenchyma (CT scan Chest)

At histopathology department we received multiple, pale yellow to grey tan soft tissue with outer hemorrhagic areas all together measuring 25 x 16 x 3 and weighing 1.1 kg. Cut surface pale white to yellow, homogeneous appearance. Random sections were given in 12 blocks.

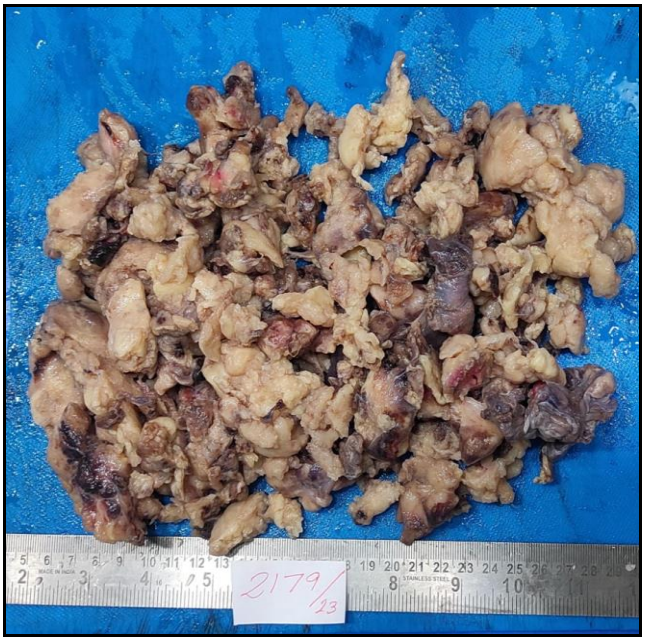

Fig 3: Gross picture showing multiple pale yellow to grey tan hemorrhagic tissue.

Microscopy showed lobules of hyperplastic thymic tissue displaying cortex, medulla with lamellated hassals corpuscle. Proportional increase in lobules of mature adipose tissue surrounded by thin fibrous capsule intimately associated with the thymic tissue noted. Thick bands of fibrous tissue with congested blood vessels and dispersed neutrophils seen dividing the thymic tissue. No evidence of malignancy. Final diagnosis of a thymolipoma was given.

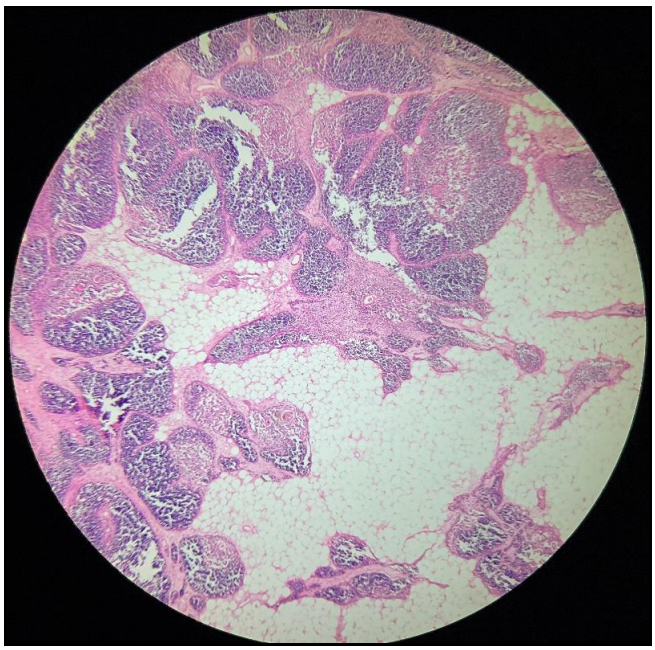

Fig 4: Low power view shows thymic tissue with admixed adipocytic tissue (Hematoxylin & Eosin 100x)

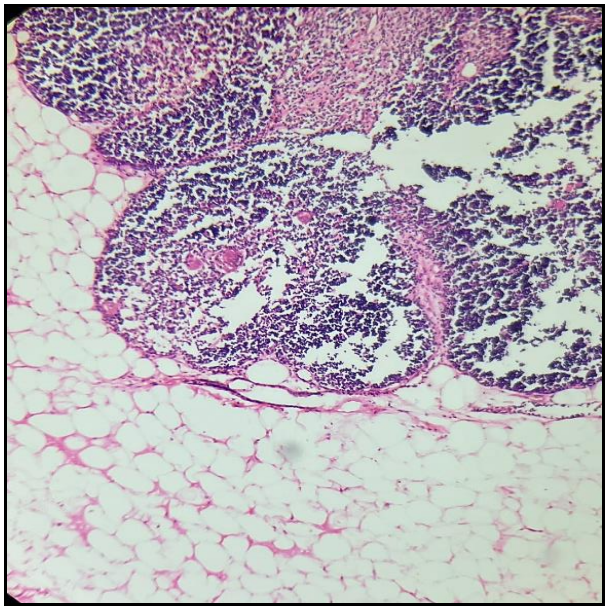

Fig 5: High power view of thymic tissue with hassals corpuscle and benign adipose tissue (Hematoxylin & Eosin 400x)

Discussion

Thymolipoma is an encapsulated tumour that consists of mature adipose tissue with interspersed non-neoplastic thymic tissue. In 1949, Hall, suggested this terminology. and proposed that this lesion is a true mixed tumor comprising of fat and thymic tissue [3]. Thymolipoma can occur at any age, has no sex prediliction and comprises about 2 to 9% of all thymic neoplasms [4]. Although this lesion is mostly asymptomatic or might be associated with pressure related symptoms, some patients may have symptoms associated to paraneoplastic syndromes, such as myasthenia gravis, graves' disease, hypogammaglobulinemia, aplastic anemia, and lichen planus [5]. Thymolipomas are classified under mesenchymal tumors in 5th Edition of World Health Organisation classification of thoracic tumors.

Thymolipomas range in size from 4 to 36 cm, and on radiograph may simulate cardiomegaly and diaphragmatic elevation. Despite clinicopathological reviews and several case reports the pathogenesis of this lesion still remains unclear. Multiple theories are postulated for the development of thymolipoma which include neoplastic theory, hamartomatous theory and a regression theory.

a) The neoplastic theory states the possibilities of a pure mesenchymal origin (lipoma of thymic fat) or a mixed endodermal and mesenchymal tumors. (both neoplastic thymic and adipose tissue).
   Maekawa *et al* [6] reported a case of a thymolipoma that was associated with increased serum levels of Carbohydrate antigen 19-9 (CA19-9). They suggested that the neoplastic growth of thymus caused increased CA19-9 production, which was confirmed by postoperative normalization of serum CA 19-9 levels.
b) Second theory suggests that thymolipomas are thymic hamartomas, formed by an admixture of white adipose tissue (30% to 80%) associated with thymus epithelium [7].
c) The regression category includes 2 theories of thymic involution. They may be as a result of involution of thymic hyperplasia which is due to fatty replacement of true thymic hyperplasia or of a regressed thymoma in which patients are mostly associated with myasthenia gravis. A thymoma may also undergo regression.

Grossly thymolipomas are yellow masses with well-defined lobulated contours and are encapsulated. They have yellow, soft, homogeneous consistency on cut surface. Areas of necrosis or hemorrhage are not generally seen. Microscopically the lesion comprises of mature adipose tissue and strands or even larger areas of thymic tissue. The adipose tissue content of thymolipomas varies, usually represents 50-85% of the lesion, however it can account for almost 95% of the lesion in some cases [8].

Few variants of thymolipoma include thymofibrolipoma and thymoangiolipoma. Thick collagen fibers present extensively in association with islands of mature adipose tissue are seen in thymofibrolipomas. In thymoangiolipoma variable proportion of mature fat tissue, thymic component, and blood vessels are noted. In some cases, as in normal thymus, myoid cells also known as striated cells may be detected in thymolipomas which are thought to be due to metaplastic change of reticuloepithelial thymic cells, or cells from neural crest origin or from perithymic mesenchymal cells incorporated into the thymus. Atypia or mitotic activity is not seen. Thymolipomas rarely may contain a neuroendocrine tumor or a thymoma. Immunohistochemistry is not necessary.

The differential diagnosis includes lipoma (usually present above the diaphragm and doesnot show thymic tissue), mediastinal lipomatosis, thymic hyperplasia (capsule, predominance of adipose tissue and peripheral rimming of normal thymus favour thymolipoma), thymoma, mature teratoma (identification of other components like sebaceous glands, cartilage, gastrointestinal and respiratory epithelium may point towards a teratoma), and malignant neoplasms such as liposarcoma(presence of lipoblasts), lymphoma, and thymic carcinoma.

Conclusion

Thymolipoma can be clinically asymptomatic. Radiological investigations with findings of fat containing structure arising from the anterior mediastinum along with internal fat stranding & nodularity can play an important role in diagnosis. Histopathology is essential for final diagnosis. Surgical excision is the mainstay of treatment.

Conflict of interest

None

Source of funding

None

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