Letters to the Editor

Post Congenital Pulmonary Airway Malformation Excision Thymic Growth: An Unusual Postoperative Sequel

Sir,

Congenital pulmonary airway malformation (CPAM) is the most common cystic lung disease of childhood. Around one-third of neonates with CPAM has respiratory distress in the immediate postnatal period. Complete resection of CPAM with lobectomy through thoracotomy or thoracoscopy remains the treatment of choice. In this report, we highlight a hitherto unreported skiagram finding of apparent thoracic mass in two of our postoperative CPAM patients.

Case A: A 9-month-old girl, a follow-up case of left lower lobe CPAM. She underwent left thoracotomy and excision of left lower lobe CPAM at 5 months of age and was discharged in the stable condition on the 5th postoperative day. The child remained asymptomatic in the postoperative period. However, at 9-month age, the child had an episode of fever and cough for 2 days when she underwent chest skiagram at another healthcare center. A radio-opaque shadow was noted in the anterior mediastinum on the chest skiagram, and the child was urgently referred to our center with suspicion of recurrence or neoplastic lesion. Thoracic ultrasonography was done for the mediastinal mass evaluation, and it was documented as enlarged thymus. No other intervention was done, and the child remained asymptomatic in the follow-up period. The thymic growth decreased in the next 8 months.

Case B: A 10-month-old boy, also a follow-up case of left upper lobe CPAM. The child underwent left thoracotomy and excision of left upper lobe CPAM at 7 months of age. The child had prolonged chest-tube drain output postoperatively with intermittent episodes of fever for two days; however, chest-tube drain was removed by postoperative day 5, and the child was discharged by the 7th postoperative day. On a 3-month postoperative follow-up visit, a history of febrile illness was elicited, and hence, a follow-up chest skiagram was advised, which demonstrated a faintly delineated opaque shadow in the mediastinum. Thoracic ultrasonogram confirmed it to be a thymic growth with the classical “starry sky” pattern of the thymus. The child remained asymptomatic till the last follow-up of 16 months with subsequent resolution of the thymic enlargement.

The postoperative thymic growth is not described specifically in the postoperative course of patients of CPAM. These cases are the first cases in the literature in which such a finding is being reported. Since the thymic enlargement resolved spontaneously, it can be attributed to nonneoplastic thymic growth. Thymic enlargement is a physiological process in infants and young children. Another differential diagnosis is thymic hyperplasia which is rarely reported in infancy. Thymic hyperplasia can be histologically classified as true and lymphoid. True thymic hyperplasia has been found associate with major surgery or burns, infection, radiation, and chemotherapy, while lymphoid thymic hyperplasia is associated with immunological or autoimmune pathology. The radiological assessment with thoracic

![Figure 1](image_url)

Figure 1: (Case a) The chest X-ray of a 5-month-old girl showing the preoperative large left lower-lobe CPAM (as white asterix) (a). Child (at 9-months of age) had a postoperative X-ray demonstrating anterior mediastinal mass (as white arrowheads) (b), which was diagnosed as thymic growth (starry-sky appearance) on chest ultrasonogram (c). (Case B) The chest X-ray of a 7-month-old boy showed preoperative large left upper-lobe CPAM (as white asterix) with mediastinal shift to right (white arrow) (d). The postoperative chest X-ray at age of 10 months demonstrated anterior mediastinal mass (as white arrowheads) (e), which was subsequently reduced by 16-months postoperative chest X-ray (f). (CPAM: Congenital Pulmonary Airway malformation)
However, sudden thymic enlargement requires surgical follow-up with proper counseling of parents, and both had a satisfactory outcome on follow-up. A residual lesion’s possibility was minimal; since residual disease is reported more after segmentectomy (15%), while none with lobectomy.[3]

The compensatory growth of the lung after resection of a diseased lung or lobectomy has been well documented.[1] However, sudden thymic enlargement after surgery followed by regression in infants is not reported. Both children in the present report had large cystic lung lesions occupying the entire hemithorax. The hypothesis behind these findings can be: (1) the surgical stress response of the thymus, (2) physiological hypertrophy of the thymus, or (3) the mediastinum shift leading to change in the alignment and possible rotation of the thymus in the vacant mediastinal space making it more apparent in a skiagram. These hypotheses can be tested or assessed by studying many operated patients with cystic lung lesions of varying sizes and presentation, although there is no existing protocol to subject such patients to routine postoperative radiographs unless clinically warranted.

However, if a chest skiagram is done in the follow-up period, which suggests an intrathoracic mass, the diagnosis of thymic growth should always be kept as a differential, and a focussed ultrasound should be helpful to sort out the diagnosis. Also, healthcare workers should be aware of these sequelae while counseling the parents of patients with CPAM. If not informed, radiographic findings like in our cases could heighten their anxiety and expose the infants to unnecessary costly investigations or radiation. A note must also be made of the Sail sign which is characteristically described as the triangular extension of the normal thymus laterally towards the right. The right lobe of the thymus has a convex lateral margin, and its straight inferior border gives the sail-like appearance. However, in our cases, the orientation was skewed possibly in view of the CCAM resection-related mediastinal shift and rotation of the thymus, thereby the classical Sail sign was not observed rather a left sided shadow was found.

In conclusion, the postoperative thymic growth in CPAM is an underreported physiological phenomenon that can be evaluated and followed up by non-invasive investigations and may not require any intervention. Awareness of this possible postoperative sequelae can alleviate the anxiety of both health-care workers and parents alike.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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