Case Report

Surgical Occlusion of Leaking Bronchus by a Serratus Anterior Flap in a Child with Langerhans Cell Histiocytosis

Lokavarapu Manoj Joshua, Ashok Rijhwani, Manish Kumar Gupta, Enono Yhoshu, Gyanendra Chaudhary

Langerhans cell histiocytosis is an uncommon disease of childhood. Intrathoracic transposition flaps have been described for a management of number of conditions. We discuss our experience of the use of serratus anterior flap for the obliteration of a pulmonary bulla with a communicating airway, in a 1½-year-old pediatric patient with multisystem Langerhans cell histiocytosis who presented with recurrent pneumothorax with empyema due to rupture of bullae.

Keywords: Bronchopleural fistula, emphysematous bullae, Langerhans cell histiocytosis, pneumothorax, serratus anterior flap

INTRODUCTION

Langerhans cell histiocytosis (LCH) is an uncommon disease of childhood with incidences varying from 2 to 9 per million per year. The peak occurrence is between 1 and 4 years of age with a male predominance. Lesions are characterized by CD1a+/CD207+ dendritic cells with inflammatory infiltrate.[1] LCH can involve either single or multiple systems, and the clinical presentation in LCH varies depending on the organ of involvement. Isolated pulmonary involvement is uncommon in children, but is common in multisystem disease. Our patient had multiple systems involved, with documented disease affecting both lungs and the liver.[2]

We discuss our experience of the use of serratus anterior flap for the obliteration of a pulmonary bulla with a communicating airway, in a patient with LCH, with multiple system involvement. This 1½-year-old child came with recurrent pneumothorax due to rupture of bullae.

CASE REPORT

A 1½-year-old male child presented with dyspnea, high-grade fever, cough without expectoration, and with yellowish discoloration of the eyes and skin. There was a history of recurrent respiratory tract infections with empyema with recurrent right-sided pneumothoraces for which intercostal tube drainage had been done in the past, on multiple occasions.

On examination, the child weighed 7.3 kg, with a length of 72 cm. He was tachypneic, febrile, icteric, and malnourished. His oxygen saturation was above 95% with oxygen supplementation by nasal prongs at 2 L/min. Routine blood tests showed of leukocytosis of 14580/mm³, with 75% being neutrophils. The total bilirubin was 11 g/dl with a direct bilirubin of 6 mg/dl. Chest X-ray revealed the presence of a large pulmonary bulla in the right hemithorax with evidence of compression of the right lung and significant mediastinal shift. Ultrasound of the abdomen showed a possible thrombus of the portal vein with a wedge-shaped possible infarction of segments 6 and 7 of the liver. This was confirmed on the magnetic resonance imaging examination of the abdomen, which also showed multiple well-defined T2 hyperintense and T1 hypointense small cystic areas involving both the lobes of the liver, the largest measuring 5 mm × 6 mm in segment 8. High-resolution computed tomography (CT) scan of the thorax showed multiple cystic lesions in both the lungs, the largest measuring 2 cm × 1.5 cm in the right lower lobe along with multiple pulmonary bullae, and hydropneumothorax of the right hemithorax [Figure 1].

Address for correspondence: Dr. Lokavarapu Manoj Joshua, Department of Surgery, All India Institute of Medical Sciences, Rishikesh, Uttarakhand, India. E-mail: manoj.josh1993@gmail.com

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Joshua LM, Rijhwani A, Gupta MK, Yhoshu E, Chaudhary G. Surgical occlusion of leaking bronchus by a serratus anterior flap in a child with Langerhans cell histiocytosis. J Indian Assoc Pediatr Surg 2021;26:123-5.
An urgent thoracoscopic partial decortication, decompression of bullae, with lung biopsy with chest tube insertion was done. Thoracoscopy findings were multiple septations in pleural cavity with bullae in the middle lobe of the right lung. The patient’s clinical condition improved and the chest X-ray showed good expansion of the lung on the 5th postoperative day, with no pulmonary bullae or air leak.

We instilled a single dose of oxytetracycline, through the intercostal tube, on the 9th postoperative day in an attempt to effect a chemical pleurodesis. We removed the intercostal 24 h later.

Lung biopsy showed tumor cells positive for CD1a and S100. The morphological and immunohistochemical features are consistent with Langerhans cell histiocytosis.

Three days after the pleurodesis, the patient developed new bullae and tachypnea. We decided to do a thoracotomy and open decortication. Multiple bullae of varying size from 2 mm to 3 mm to 2 cm involving the right middle and lower lobes were present. A parietal pleurectomy was done and the pulmonary bullae deroofed. One significant bulla, situated in the major fissure, had a large airway entering the floor of the bulla, causing a persistent air leak [Figure 1]. This airway was closed using vicryl suture and the closure was buttressed by a muscle flap consisting of the fifth digitation of the right serratus anterior muscle. This digitation was detached from the anterior origin, preserving its posteriorly based blood supply. Two intercostal drains kept postoperatively which were removed on day 5 and day 10 after surgery. The child was stable, ventilating well in air and feeding normally from the 2nd postoperative day. There was good lung expansion with no evidence of air leak [Figure 2]. Chemotherapy for the LCH was being planned.

**DISCUSSION**

The clinical manifestations of pulmonary involvement in LCH include cough, dyspnea, chest pain, and other constitutional symptoms. Recurrent pneumothorax (unilateral or bilateral) may develop during the course of illness. High-resolution CT scan of the chest is a good modality to detect pulmonary involvement, which shows cystic or nodular lesions. The diagnosis is best established by lung biopsy with immunohistochemical staining for CD1a. The diagnosis is established by lung biopsy in our case.[3]

Our patient presented with empyema with pneumothorax, which is uncommon presentation. The patient has a history of recurrent respiratory tract infections in the past and tube thoracostomies done for recurrent pneumothorax. Our initial management included a thoracoscopic-guided decortication and decompression of the bulla and a lung biopsy. However, there was a recurrence of the bulla in 10 days’ time and therefore we decided to do an open exploration. We found a significant airway leaking in the floor of the bulla, and this led us to using a digitation of the serratus anterior muscle to close the leak.

Intrathoracic transposition flaps have been described for several conditions such as bronchopleural fistula, space obliteration for postpneumonectomy empyema, tuberculosis empyema, buttressing of bronchial and tracheal anastomosis, and repair of tracheoesophageal fistulas. Commonly used muscle flaps are those from the latissimus dorsi, intercostal muscles, serratus anterior, and transversus/rectus abdominis.[4,5]

In our patient, we describe the use of a serratus anterior flap for obliteration and buttressing of cyst cavity and the airway leak in a child with LCH, which had not been described before. This is to help in the treatment of the rupture bullae, till he can undergo definitive treatment which includes chemotherapy and a possible lung transplant.
Serratus anterior muscle flap is a good choice due to its reliable and segmental blood supply by thoracodorsal artery, easy surgical access when used as a pure muscular flap, long arc of rotation, limited mutilation, and less impact on respiratory function.

The postoperative complications include shoulder instability with functional impairment at abduction and elevation over 120 degree, winging or asymmetry of scapula, seromas, bleeding, and flap necrosis.[4,5]

In our case, a single digitation was used which did not cause any scapular deformity or abnormal shoulder movement.

**CONCLUSION**

The serratus anterior muscle flap is a versatile flap which can be used to repair bronchial air leaks causing pneumothorax, emphysematous bullae, and bronchopleural fistulae. It can be used with good effect in a variety of clinical settings, acute, subacute, and chronic.

**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 initial s will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**References**

1. Abla O, Egeler RM, Weitzman S. Langerhans cell histiocytosis: Current concepts and treatments. Cancer Treatment Rev 2010;36:354-9.

2. Haupt R, Minkov M, Astigarraga I, Schäfer E, Nanduri V, Jubran R, et al. Langerhans cell histiocytosis (LCH): Guidelines for diagnosis, clinical work-up, and treatment for patients till the age of 18 years. Pediatr Blood Cancer 2013;60:175-84.

3. Jezierska M, Stefanowicz J, Romanowicz G, Kosiak W, Lange M. Langerhans Cell Histiocytosis in Children – A Disease with Many Faces. Recent Advances in Pathogenesis, Diagnosticexaminations and Treatment. Vol. 35. Postepy Dermatologii i Alergologii: Termedia Publishing House Ltd.; 2018. p. 6-17.

4. Widmer MK, Krueger T, Lardinois D, Banic A, Ris HB. A comparative evaluation of intrathoracic latissimus dorsi and serratus anterior muscle transposition*. Eur J Cardio-Thoracic Surg 2000;18:435-9.

5. Botianu PV, Botianu AM, Dobrica AC, Bacarea V. Intrathoracic transposition of the serratus anterior muscle flap–personal experience with 65 consecutive patients. Eur J Cardiothorac Surg 2010;38:669-73.