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Carinal resection and reconstruction following inflammatory myofibroblastic tumor resection: A case report

Julia G. Lyon [a,*], Lauren A. Kilpatrick [a,1], Tara L. Rosenberg [a,1], Abby R. Nolder [a,1], Michiaki Imamura [b], Gresham T. Richter [a,1]

[a] Department of Otolaryngology, University of Arkansas for Medical Sciences, 4301 West Markham Street, Slot 543, Little Rock, AR 72202, USA
[b] Department of Cardiothoracic Surgery, University of Arkansas for Medical Sciences, Little Rock, AR, USA

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ABSTRACT

Inflammatory myofibroblastic tumors (IMT) are rare tumors of the respiratory tract that most commonly occur in the lung and are rarely seen in the trachea. They present most often in young patients. We report on a case of an IMT of the carina in a seven year old girl, requiring carinal resection and reconstruction with a novel technique in pediatric airway surgery. Attempts at endoscopic excision of the carinal IMT were unsuccessful. An open approach for resection of the involved carina, distal trachea, and proximal mainstem bronchi was performed via sternotomy and cardiopulmonary bypass. The resulting triangular defect in the trachea and bronchi was reconstructed with anastomosis of the proximal trachea and left mainstem bronchus using a rotational flap of the right lateral mainstem bronchial wall. The remaining right mainstem bronchus was anastomosed, end to side, to the intact trachea proximal to the primary anastomosis. Bronchoscopy and MRI 22 months post resection and reconstruction revealed a healthy neo-carina and patent distal airway with no evidence of recurrent IMT. Pediatric patients with carinal inflammatory myofibroblastic tumors can be successfully managed with open resection and reconstruction of the airway.

Inflammatory myofibroblastic tumors (IMT) are rare tumors that most commonly are found in the lung and rarely occur in the airway. This is the report of a 7 year old girl with an IMT involving the carina. Attempted resections by endoscopy were unsuccessful. She was then treated with open resection of the carinal tumor that included the distal trachea and proximal mainstem bronchi via sternotomy and cardiopulmonary bypass. Airway reconstruction reanastomosed the main bronchi to the trachea forming a neo-carina. MRI and bronchoscopy showed no evidence of recurrence 22 months post-operative.

1. Case report

A 7 year old girl who presented with cough, wheezing, and pneumonia was found to have a carinal mass causing nearly complete obstruction of the trachea (shown in Fig. 1a).

Biopsy showed inflammatory myofibroblastic tumor. The tumor was debulked via endoscopy on three separate occasions but recurred. Total resection via sternotomy and cardiopulmonary biopsy was decided to attempt cure. Pre-operative bronchoscopy revealed that the tumor extended from the carina up the posterior tracheal wall and partially obstructed the left main bronchus.

1.1. Open resection of the carinal IMT

The child was intubated, sternotomy performed, and the child placed on cardiopulmonary bypass. The carina and mainstem bronchi were dissected. The tumor was noted to extend through the tracheal wall, but did not appear to involve any mediastinal structures. The tumor was removed in bloc and resection extended until all margins were negative. Multiple paratracheal and carinal lymph nodes were removed and all were negative for tumor.

1.2. Airway reconstruction

Airway reconstruction was then undertaken. The defect in the trachea and bronchi was reconstructed with anastomosis of the

* Corresponding author. Tel.: +1 501 364 1047; fax: +1 501 364 4790.
E-mail address: jlyon@uams.edu (J.G. Lyon).
[1] Tel.: +1 501 364 1047; fax: +1 501 364 4790.

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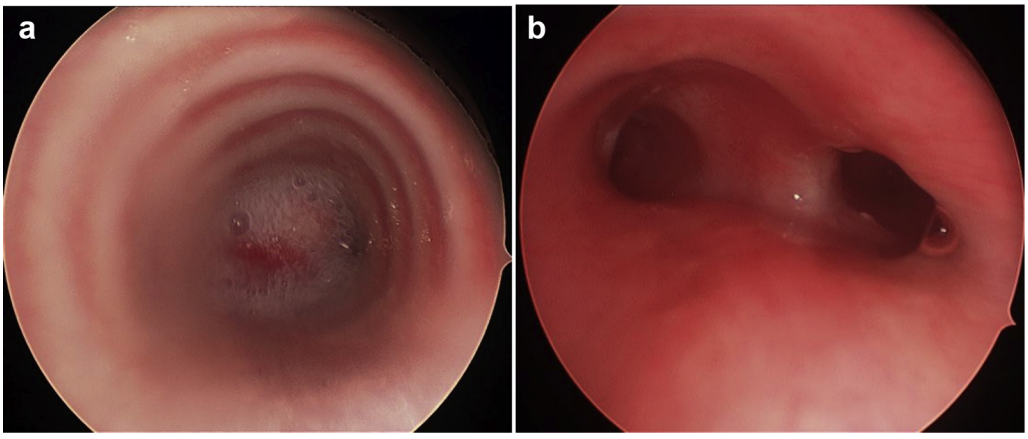

Fig. 1. Bronchoscopy at initial presentation (a), and 5 months post-operative (b).

proximal trachea and left mainstem bronchus using a rotational flap of the right lateral mainstem bronchial wall (see Fig. 2a). This created a continuous conduit between the trachea and left main bronchus. Some buckling was repaired; no leaks were evident.

An end to side anastomosis was planned to rejoin the right main bronchus to the trachea as shown in Fig. 2b. An oval excision of the lateral tracheal wall, about 2 cm away from the left mainstem/trachea anastomosis and anterior to the trachealis, created an opening. The right mainstem was anastomosed to the opening in the tracheal wall. A small leak was observed and repaired with 4-0 PDS suture.

The left mainstem was noted at this time not to be ventilating well. Intraoperative bronchoscopy revealed buckling of the lateral bronchial wall which was resected and repaired. Subsequent positive pressure showed no leaks at all repair sites with good ventilation of both lungs.

Stay sutures were used to secure the anastomoses. Fibrin sealant was applied.

1.3. Post-operative course

The child was removed from bypass, the surgical site closed, and the child taken to the ICU intubated. She was extubated 3 days later, weaned off of oxygen over the subsequent 3 days and discharged.

Serial bronchoscopies at increasing time intervals revealed healthy repair sites and anastomoses. Fig. 1b above shows the healthy neocarina 5 months post-operatively. Breath sounds were equal bilaterally throughout her recovery. At 22 months follow up the child has complete resolution of presenting symptoms with no evidence of tumor recurrence by MRI (Fig. 3) and endoscopy. She denies any airway symptoms and continues normal physical activity.

2. Discussion

IMT are rare tumors that most generally present in the lung, but are known to rarely occur in the trachea or bronchi and can occur in the carina [1]. Lung parenchymal IMT are usually asymmtomatic; however, when located in the airway the tumors often present with cough, dyspnea, or pneumonia.

These tumors are usually considered benign, but metastasis has been described [2] and subtotal resection usually results in recurrence of the tumor. Complete resection is the recommended treatment [3,4].

Carinal resection and airway reconstruction has been demonstrated to be a successful treatment for carinal tumors [5]. In children, special attention should be paid to preventing stenosis of

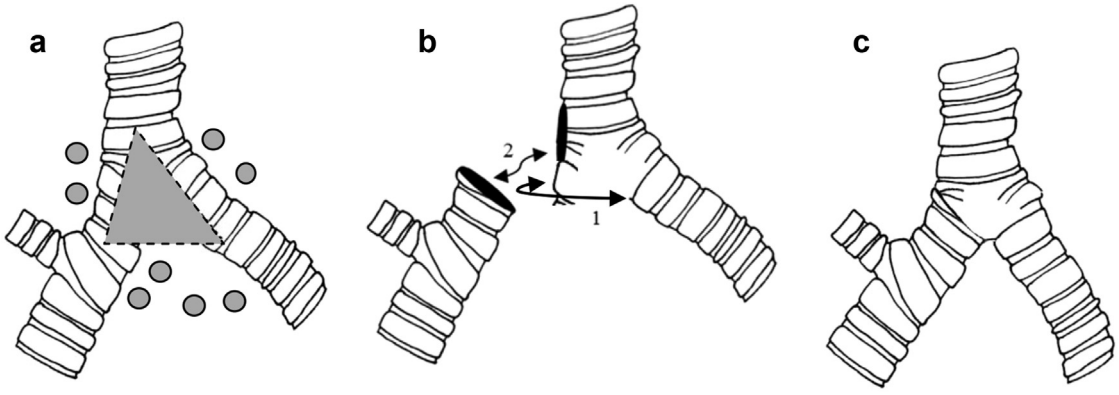

Fig. 2. Airway Reconstruction, removal of tumor and lymph nodes (a), rotational flap anastomosis of proximal trachea and left mainstem (b, arrow 1) and end to side anastomosis of right mainstem to trachea (b, arrow 2), complete repair (c).

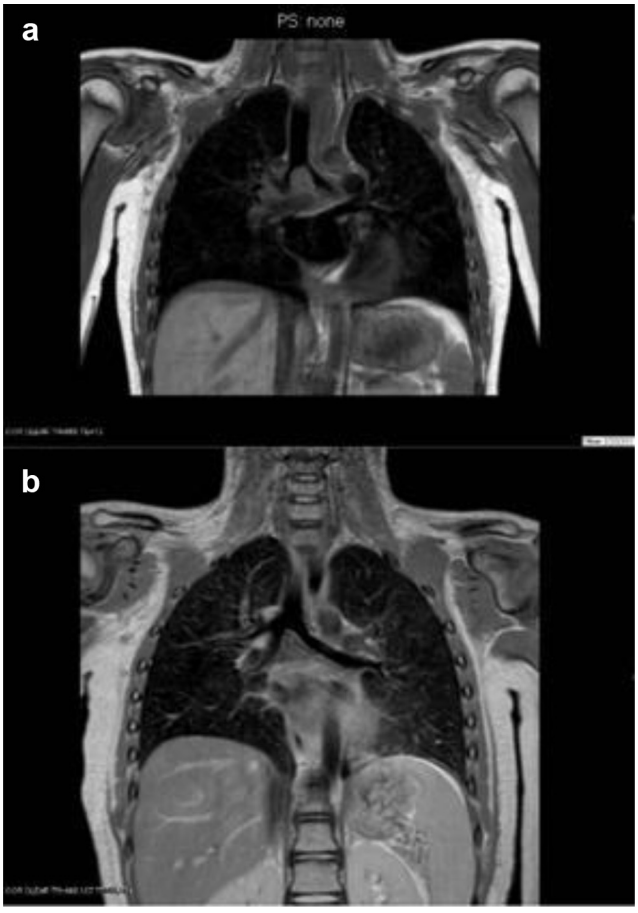

Fig. 3. MRI pre-operative (a), and 22 months post-operative (b).

an already small airway and using dissolvable sutures in the anticipation of further growth of the child [6].

Preserving the carinal shape in the repair is beneficial and the elasticity present in children's airways is more amenable to the creation of a neocarina than adult airways [6].

Here we demonstrate a case of total resection with complex distal trachea, carina and bronchial reconstruction using a bronchial flap. In this case, the tumor margins and mediastinal lymph nodes were negative for tumor. This should reduce the risk of recurrence of the IMT, however close observation will be maintained to evaluate for recurrence or metastasis. This patient remains symptom free with a patent neocarina more than 22 months post open resection of her carinal tumor and airway reconstruction.

3. Conclusion

Pediatric patients with carinal inflammatory myofibroblastic tumors can be successfully managed with open resection and reconstruction of the distal airway.

Conflicts of interest

None.

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