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Use of serial rigid bronchoscopy in the treatment of plastic bronchitis in children

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ABSTRACT

Aim: Plastic bronchitis (PB) is a rare disorder characterized by formation of bronchial casts (BC) in the tracheobronchial tree with partial or complete airway obstruction. Although lysis of casts with several fibrinolytic agents has been reported, removal of BC with bronchoscope provides better clearance of airways. A retrospective study was performed to evaluate the use of serial rigid bronchoscopy (RB) in the treatment of PB in children.

Patients and methods: Between 2011 and 2015, children with partial or complete airway obstruction with PB were evaluated for age, gender, underlying disease, clinical findings, results of bronchoscopic interventions and histopathologic findings.

Results: Five patients with 14 RB interventions were evaluated. The mean age of the patients was 7.8 years (min: 3 years — max: 14 years) and male–female ratio was 4:1. All of the patients were diagnosed as asthma and none of them had underlying cardiac disease. Suction of mucus plaques and bronchoalveolar lavage were performed in all patients with flexible bronchoscopy. Also, aerosolized tissue plasminogen activator was used in two patients. During follow-up serial RB was indicated in patients with persistent atelectasis and severe airway obstruction. The most common localization of BC was left main stem bronchus and bilateral cast formation was detected in 7 interventions. Although, removal of BC was challenging in two patients because of cast friability and fragmentation, most of the plugs were successfully removed with optical forceps and rigid suctioning. Two patients underwent repeated RB (min: 3 — max: 8) for recurrent symptoms. Histopathologic evaluation of BC revealed Charcot–Leyden crystals with inflammatory cells in all patients. The time interval between RB interventions was one to five months.

Conclusion: BC are tenacious mucus plugs which are firmly wedged to the tracheobronchial tree. The use of optical forceps with rigid suction provides adequate removal of BC during RB. Because of underlying disease, it is difficult to cure cast formation. Therefore, most of the patients require serial RB when they become unresponsive to standard therapy or develop partial or complete airway obstruction.

Plastic bronchitis (PB) or cast bronchitis is a rare condition characterized by presence of mucofibrinous plugs that can partially or completely obstruct the tracheobronchial tree [1]. Although the pathophysiology of PB formation is not well understood, children with underlying cardiac disease are at risk of developing bronchial casts especially after surgical procedures that direct systemic blood flow into the pulmonary circulation [2]. Casts are more adhesive than mucous plugs, and conform the shape of the tracheobronchial tree [3]. Therefore, it may completely occlude the airway and present with cough, fever, dyspnea and wheezing.

Several therapeutic agents including inhaled or systemic corticosteroids, aerosolized acetylcysteine, tissue plasminogen activator (tPA), and intravenous and aerosolized heparin have been used for PB [4]. Patients who are unresponsive to medical treatment and profound airway obstruction may require flexible and rigid bronchoscopic lavage and direct bronchoscopic removal of casts [5]. However, none of them has been proven to be effective for all patients. In spite of promising results with fibrinolytic agents, BC bronchial cast usually recurs because of underlying disease. Therefore, removal of BC with bronchoscopy provides better clearance of airway. A retrospective study was performed to evaluate the use of serial rigid bronchoscopy in treatment of PB in children.

1. Patients and methods

Between 2011 and 2015, children with partial or complete airway obstruction with PB were evaluated for age, sex, underlying disease,

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Table 1
Clinical features and flexible bronchoscopic findings of patients.

	Case 1	Case 2	Case 3	Case 4	Case 5
Sex/age (years)	M/7	F/10	M/5	M/3	M/14
Initial symptom	Recurrent lung infections	Asthma, left lower lobe atelectasis	Recurrent lung infections	Recurrent lung infections	Asthma, recurrent lung infections, left lung atelectasis
Physical examination	Decreased chest sounds on left hemithorax	Decreased chest sounds on left lower lobe	Decreased chest sounds on left hemithorax	Fine rales on right lung and bilateral bronchi	Bilateral fine rales, lymphedema on left legs
Chest X-ray	Left lower lobe atelectasis	Left lower lobe atelectasis	Left lung total atelectasis	Left lung total atelectasis	Interstitial infiltration
Chest CT	Left lower lobe subsegmental atelectasis	Left lower lobe subsegmental atelectasis	Left lung total atelectasis	Mediastinal LAP (1.5 cm size maximum) Left upper lobe atelectasis, Left lower lobe bronchial dilatation, Right lower lobe and middle lobe atelectasis	Left lung total atelectasis
Echocardiography	Normal	Normal	Normal	PDA	Normal
Previous asthma history	No	Yes	No	No	Yes
Lung biopsy					Lipid laden macrophages
Flexible bronchoscopy	Bronchial cast in left upper lobe and lower lobe bronchus	Bronchial cast in left lower lobe bronchus	Bronchial cast in left main bronchus	Bronchial cast in left main bronchus and right upper and lower lobe bronchus	Bronchial cast in left main bronchus
tPA with Bronchoscopy	No	No	No	Yes	Yes
Inhaled tPA (Only at hospital)	No	No	Yes (7 days)	Yes (12 days)	Yes (10 days)
Complication	No	No	No	Yes (hemorrhage during the procedure)	No

clinical findings, results of bronchoscopic interventions and histopathologic findings.

Flexible and rigid bronchoscopic evaluations were performed under general anesthesia. Optical forceps (Storz^{R}, Germany) and rigid suctions were used during removal of casts. Histopathologic evaluations of bronchial casts were evaluated with hematoxylin–eosin staining and Charcot–Leyden crystals were considered as typical histopathologic finding for PB. Cystic fibrosis was excluded with sweat test. Immune deficiency and tuberculosis were excluded after laboratory examinations. Congenital heart diseases were investigated with echocardiography. Chest CT was performed to all of the patients.

2. Results

Five patients with 14 rigid bronchoscopic (RB) interventions were evaluated. The mean age of the patients was 7.8 years (min: 3 years — max: 14 years) and male–female ratio was 4:1. Initial symptoms, physical findings, and radiologic evaluations of patients were listed in Table 1. None of the patients had underlying cardiac disease and only two of them had a previous history of asthma. All of the patients underwent flexible bronchoscopy (FB). The findings of FB were listed in Table 1. Two of the patients received tissue plasminogen activator (tPA) during FB and three of them received inhaled tPA with a duration

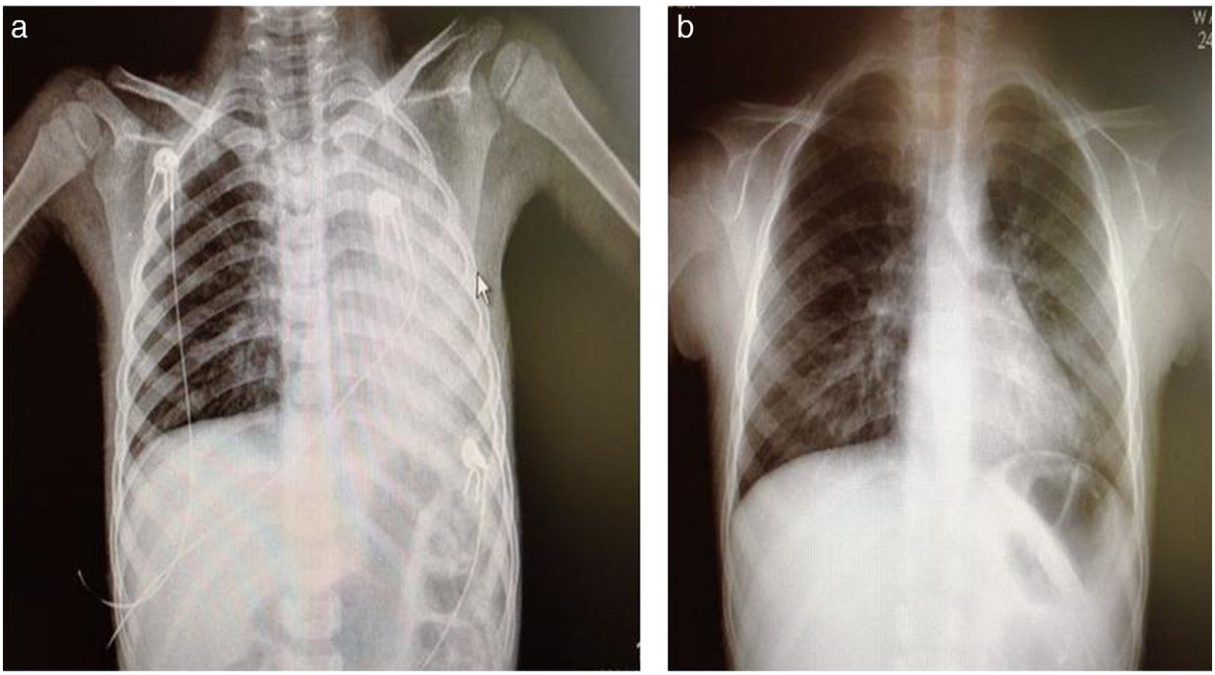

Fig. 1. Chest X-ray before (a) and after (b) rigid bronchoscopy (Case 5).