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Case Report

Indirect management of full-thickness tracheal erosion in a complex pediatric patient☆

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ABSTRACT

Prolonged tracheostomy dependence in pediatric patients can be associated with significant complications, including damage to the tracheal wall requiring reconstruction. We present a case of an 8 year-old female with full-thickness tracheal erosion secondary to the presence of a tracheostomy tube combined with a narrow thoracic inlet. A direct tracheal reconstruction was considered but eliminated due to the poor tissue quality of the trachea. Instead, a multi-disciplinary surgical team conceived of a novel indirect approach to manage the patient's tracheal defect. To our knowledge the use of indirect repair of a full-thickness tracheal defect has not been reported in the literature.

1. Introduction

Long-term tracheostomy placement is a life-saving intervention for pediatric patients with complex airway abnormalities or recurrent respiratory challenges [1]. This intervention, however, is associated with frequent complications, particularly in chronically tracheostomy-dependent patients. Here we present a case of an 8 year-old girl experiencing full-thickness tracheal erosion secondary to irritation by her tracheostomy tube in the setting of a narrowed thoracic inlet and upper mediastinum. This case presented a dilemma, as tracheal repair for a full-thickness defect carries its own risks and requires tissue of reasonable quality for anastomosis. Additionally, there was concern that the reconstructed trachea would remain compressed. Using a multidisciplinary approach in conjunction with advanced three-dimensional (3D) modeling, our team conceived an alternate indirect surgical approach to manage the tracheal defect and restore a safe airway.

1.1. Case presentation

An 8-year-old female patient with a history of mitochondrial myopathy, 2q11.2 duplication, and tracheostomy dependence presented to the ENT clinic with worsening respiratory symptoms, recurrent tracheitis, and pain. A contrast-enhanced chest CT scan revealed a narrowed thoracic inlet secondary to hypokyphosis of the thoracic spine (Fig. 1). This narrowing caused compression of the trachea between the innominate artery and thoracic spine, forcing the tracheostomy tube against the posterior-lateral tracheal wall. Bronchoscopy revealed florid intraluminal granulation overlying posterior-lateral tracheal wall at the contact point between the tracheostomy tube and tracheal wall (Fig. 2), as well as some mucosal erosion over the common wall between the trachea and esophagus. Review of the CT images demonstrated a large mass of extra-tracheal granulation, suggesting a full-thickness erosion of the airway at the level of the tracheostomy tube. The granulation tissue filled the posterior aspect of the trachea, extending beyond the tracheal walls into the posterior mediastinum. The presence of this granulation tissue prevented precise measurement of the tracheal wall defect on endoscopy or imaging. However, we suspected that the region of tracheal erosion measured roughly 6 × 16mm, based on the size of the granulation tissue immediately adjacent to it in the mediastinum. The patient did not show clinical signs of mediastinitis; our supposition is that the extra-tracheal granulation served to seal off any major areas of secretion leakage through the tracheal defect. To supplement radiographic and bronchoscopic workup, a 3D model of the musculoskeletal, cardiovascular, and tracheal anatomy of the patient was printed using methods previously described by our group [2] (Fig. 3). In addition to confirming the previous findings, the model clearly demonstrated an additional anatomic restriction caused by posterior subluxation of the left clavicular head which was not appreciated on 2D images.

Topical therapy and alternate tube sizing was trialed without

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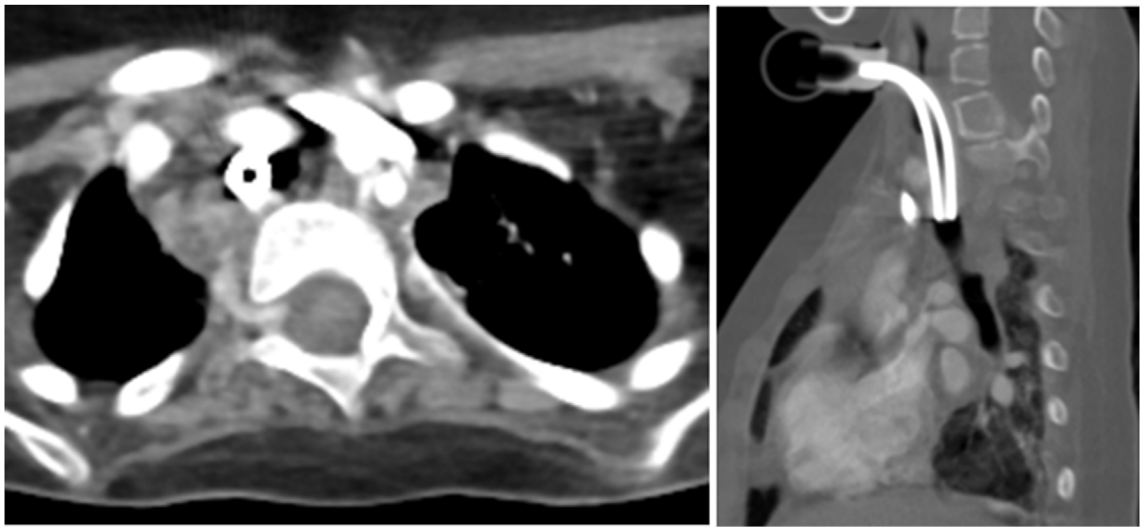

Fig. 1. Contrast-enhanced chest CT reveals hypokyphotic spine resulting in a narrowed thoracic inlet.

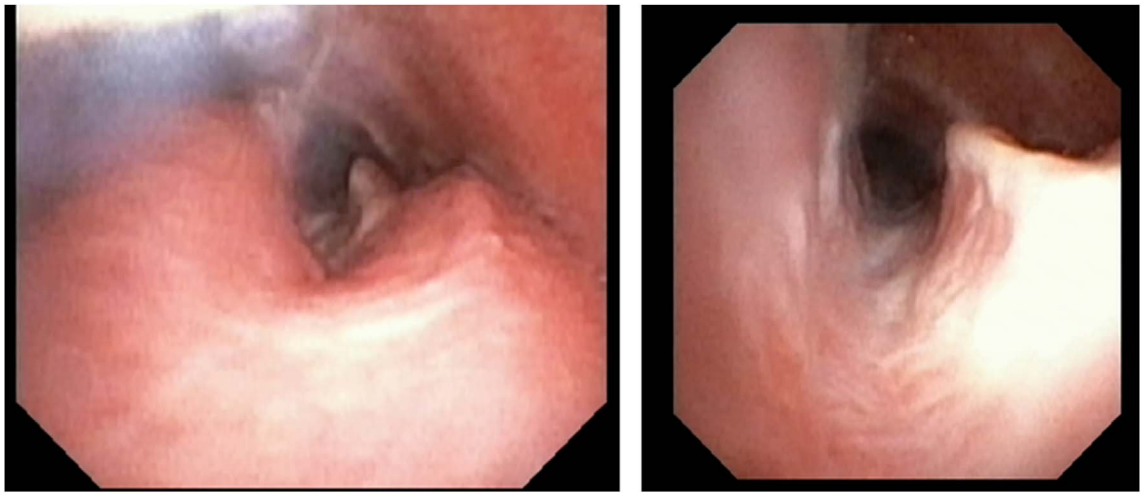

Fig. 2. Fiberoptic bronchoscopy reveals posterolateral tracheal erosion with granulation tissue.

success in alleviating the tracheal irritation and granulation. Secondary to the potential catastrophic consequences associated with a complete perforation of the tracheal wall or development of a tracheoesophageal fistula, definitive operative management was determined to be the best alternative. A multi-disciplinary surgical team was formed including: pulmonology, otolaryngology, cardiovascular surgery, general surgery, and orthopedic surgery. A direct reconstruction of the trachea was considered, but given the friability of the damaged tissue this approach would be potentially unsafe and in any case would not definitively address the compression of the trachea. A tracheal homograft was not commercially available as a salvage option. Posterior approach spine reconstruction to restore kyphosis was considered, but deemed unlikely to succeed due to the likelihood of ongoing progressive spine deformity. Instead, the multi-disciplinary team conceived a novel surgical procedure to repair the trachea through an indirect approach. This approach would decompress the anterior mediastinum allowing for the tracheostomy tube to lift off the tracheal wall while avoiding direct intervention at the site of tracheal erosion. The procedure included: (i) bony decompression including complete manubriectomy, bilateral resection of clavicular heads and first ribs (ii) partial thymectomy, and (iii) re-implantation of innominate artery with interposition graft to minimize tension (Fig. 4). Removal of the clavicular heads was acceptable in this patient because she did not use her arms for any weight-bearing activities. Given her ventilator dependence and ability to trigger with her diaphragm resection of the first ribs was also included.

The procedure was completed with no intra or perioperative complications. Postoperatively, she was noted to have unilateral phrenic nerve weakness. Four months postoperatively the patient's tracheal irritation greatly was improved, with minimal granulation tissue, less thickening, and no deep tracheal erosions (Fig. 5b–d). Repeat CT imaging shows complete resolution of the extratracheal granulation. Her respiratory symptoms and pain were improved.

2. Discussion

Long-term tracheostomy dependence in pediatric patients can carry a high risk of complications, especially in children with chronic or progressive underlying diseases [3,4]. In some cases, long-term tracheostomy dependence can result in tracheal irritation from the tracheostomy tube, subsequently eroding the tracheal wall and possibly fully perforating the trachea. Furthermore, the attenuated tracheal wall may be difficult to repair directly due to friability and poor perfusion. This case presents the successful management of full-thickness tracheal erosion in an anatomically complex tracheostomy dependent pediatric patient using the indirect approach of decompressing the thoracic inlet and anterior mediastinum and allowing the tracheal defect to heal by