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Successful Treatment of Severe Acquired Tracheomalacia with a Patient-Specific 3D-Printed Permanent Tracheal Splint

Robert J. Morrison, M.D.¹, Shayan Sengupta, B.S.², Colleen L. Flanagan, M.S.E.³, Richard G. Ohye, MD⁴, Scott J. Hollister, Ph.D³, and Glenn E. Green, M.D.^{5,*}

¹Department of Otolaryngology-Head & Neck Surgery, University of Michigan, Ann Arbor, MI, USA

²University of Michigan Medical School, University of Michigan, Ann Arbor, MI, USA

³Department of Biomedical Engineering, University of Michigan, Ann Arbor, MI, USA

⁴Department of Cardiac Surgery, University of Michigan, Ann Arbor, MI, USA

⁵Department of Otolaryngology-Head & Neck Surgery, Division of Pediatric Otolaryngology, University of Michigan, Ann Arbor, MI, USA

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To the Editor

Tracheobronchomalacia (TBM) is a disease of excessive collapse of the primary airways resulting from intrinsic weakness or extrinsic compression. While infantile TBM typically regresses in severity over time, adult-phenotype TBM is more often persistent and progressive¹. Severe TBM carries significant morbidity and mortality, and interventions such as surgical excision, stenting, and tracheotomy have all be associated with life-threatening complications^{2–3}.

Our group has previously had success treating infantile TBM using a bioresorbable 3D-printed external splint^{4–5}. This device has limitations in acquired disease given its' temporary design. Herein we describe a patient-specific 3D-printed external tracheal splint for treatment of TBM with modifications to device design and biomaterial selection yielding a biopermanent option for patients with adult-phenotype TBM.

*Corresponding author. Address: 1540 E Hospital Dr, SPC 5-702, Ann Arbor, MI 48109, USA. Telephone: +1 734 936 4934. Fax: +1 734 763 7802. gegreen@med.umich.edu.

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Competing Interests: The device was provided free of charge by Oxford Performance Materials. Drs. Hollister and Green are co-inventors on a United States patent for the device. The authors declare no other competing financial interests.

Methods

The patient was a 14-year-old female with severe autism and thoracic scoliosis found to have acquired tracheomalacia due to compression between a high-riding innominate artery and the thoracic spine within a narrowed thoracic inlet. Her disease progressed such that she suffered repeated cardiopulmonary arrests despite pharmacologic paralysis, mechanical ventilation, and continuous sedation for over 40 days. Previous attempts at treatment included manubriectomy, aortopexy, and innominate artery reimplantation. Endoscopic examination demonstrated continued severe, focal compressive tracheomalacia (Figure 1A, left). Withdrawal of therapy was recommended by her physicians.

We hypothesized an external splint would be effective in treating her acquired TBM. Such a device must provide long term stiffness to maintain airway patency while avoiding fatigue failure due to repetitive motion forces. Design modifications compared to the previously utilized polycaprolactone (PCL) splints are summarized in Table 1A and included removal of the bellowed topography to diffuse contact forces, thinning of the wall structure to lighten the device, and increasing the open angle of the splint to facilitate placement (Figure 1B).

Clearance for use of the device was obtained from the University of Michigan Institutional Review Board and the U.S. Food and Drug Administration. Using the patient's computed tomography (CT) scan imported into computer-aided design software (Mimics, Materialise), a model of the patient's respiratory, skeletal, and vascular anatomy was generated (Figure 1C–D). Splint geometric design parameters were determined from measurements of the affected tracheal segment and used to generate the device design (MATLAB, Mathworks). The splint stereolithography (.STL) model was digitally fit over the airway for design validation. The device was 3D-printed using polyetherketoneketone (PEKK, OXPEKK®, Oxford Performance Materials, Figure 1E). PEKK was chosen given its' superior material properties over PCL for a biopersistent implant, while also preserving the ability to manufacture the device via 3D printing (Table 1B).

Implantation was performed via a midline sternotomy approach by placing the splint around the affected trachea and suspending the trachea within the splint using polypropylene sutures (Figure 1F). Repeat endoscopy and CT imaging of the airways was performed at 1 and 12 months post-operatively. Magnetic resonance imaging (MRI) was performed at 12 months post-operatively to assess device position.

Results

Endoscopic examination immediately after splint placement showed restoration of tracheal patency (Figure 1A, right). The patient was extubated three weeks after surgery and discharged by one month. Follow-up CT imaging demonstrated complete patency of the affected trachea (Figure 1G). The hydraulic diameter of the affected region increased from $9.0 \pm 1.4 \text{ mm}^2$ to $12.1 \pm 1.0 \text{ mm}^2$ ($p < 0.001$, two-sided student's t-test, $\alpha = 0.05$). MRI demonstrated the device remained properly positioned one year after surgery (Figure 1H). The patient remains asymptomatic, has had no additional hospitalizations, and there have been no apparent complications.

Discussion

We demonstrate successful treatment of life-threatening acquired tracheomalacia with a 3D-printed biopermanent tracheal splint in a patient for whom all prior therapeutic approaches had failed. This represents a potential novel treatment for patients with severe adult-phenotype TBM not amenable to conventional therapies.

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Figure 1. Assessment, Design, Manufacturing, and Use of a 3D-Printed Polyetherketoneketone Tracheal Splint for Treating Focal Tracheomalacia

(A) Flexible bronchoscopic view of the affected trachea pre-operative [left] and immediately post-operatively [right]. (B) Comparison of design characteristics of biopermanent polyetherketoneketone [top] and bioresorbable polycaprolactone [bottom] tracheal splints. (C) Segmentation of the CT scan using thresholding function to generate patient CAD model. (D) Pre-operative CAD model showing relationship of the spine, airway, vasculature (white asterisk), and manubrium. Area of malacia denoted by black arrow. (E) 3D-printed

patient-specific polyetherketoneketone tracheal splint. **(C)** The innominate artery graft compressing the trachea [top]. 3D-printed patient-specific splint implanted onto the patient's trachea [bottom]. **(G)** CAD model of patient's trachea pre- [left] and post-operatively [right] demonstrating statistically significant increase in hydraulic diameter of the affected segment. **(H)** Patient CT [left] and MRI [right] imaging one year after implantation. Black arrow denotes location of the 3D-printed polyetherketoneketone splint.

Comparison of Device Design Characteristics and Biomaterial Properties of 3D-Printed Tracheal Splints

Table 1

A. Comparison of Upper Boundaries of Device Design Characteristics						
Device Type	Bellow Height (%)	Opening Wedge Angle (°)	Wall Thickness (mm)	Suture Hole Size (mm)	Suture Hole Spacing	Edges of Device
Bioresorbable Splint	40	90	3	1×3	Linear	Straight
Biopermanent Splint	0	120	1.5	2×2	Staggered	Rounded
B. Comparison of Biomaterial Properties						
Material	Biocompatible		Time to Resorption (years)	Young's Modulus of Elasticity (MPa)	Tensile Strength (MPa)	Surgically Modifiable
Polycaprolactone	Yes		2–3 years	350	10	Yes
Polyetherketoneketone	Yes		Never	3,000	70	No