Case Report

Nasal Reconstruction Using a Customized Three-Dimensional-Printed Stent for Congenital Arhinia: Three-Year Follow-up

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A male Mongolian child with a complete congenital absence of both nose and nasal passage had a poor survival prognosis due to respiratory distress. To enable his survival, a new nose capable of conferring respiratory function was constructed. Following reconstructive surgery, an absence of mucoperiosteum in the nasal passage can lead to rhinostenosis. To avoid this complication, a custom-made nasal silicone stent was created using three-dimensional (3D) printing technology in conjunction with the patient’s computed tomography data. The stent was implanted for 2 months to maintain the shape and size of the nasal passage. At 2 months after stent implantation, the mucoperiosteum tissue in the passage had successfully regenerated with no immune reaction. Three years after stent removal, respiratory function, nasal passage structure, and external nose shape were maintained without additional medical care. These results indicate the successful nasal reconstruction in an arhinia patient using a customized, 3D-printed nasal stent.

Key Words: Arhinia, three-dimensional printing, nasal construction, custom design.

INTRODUCTION

Arhinia, or congenital absence of the nose, is an extremely rare condition with no known cause.1–3 Failure to develop nasal placodes between the third and tenth weeks of intrauterine life can lead to congenital absence of the nose.4 Most arhinia patients have limited survival due to fetal respiratory failure during feeding and sleeping. They also suffer from respiratory inflammatory diseases, such as pneumonia and bronchitis. However, due to its rarity, nasal construction for arhinia still presents a difficult surgical challenge, as optimal techniques and timings for this procedure remain unclear.

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CASE REPORT

Patient

A 6-year-old Mongolian male exhibited complete absence of the nose and nasal cavities and had no other anomaly (Fig. 1). The site of the absent nose was flat and firm on palpation; thus, the patient could only breathe through his mouth, which had to be kept open with an instrument during sleeping to prevent apnea. He was also unable to eat while inhaling.2,5,6 To improve his quality of life and to provide him with a safe route of respiration, the surgical team decided to construct an external nose and open a new nasal passage lined with new and functional respiratory epithelium.

Surgical Processes for Nose Construction

The surgical process underlying the nasal construction was divided into two stages. First, tissue expanders were inserted into forehead tissue to obtain sufficient skin and subcutaneous tissue.7 The tissue expander was expanded gradually by periodic injection of normal saline solution over a period of 2 months.8 During the second operation, a new external nose was constructed using forehead skin flaps, subcutaneous tissue, and rib fragments to support the body of the nose. We made an inverted U-shaped incision in the skin of the new nasal area, thereby creating an inferior-based flap.3 We created the nasal passage using an electric drill guided by a navigation system (Fusion ENT Navigation System, Medtronic, MN) to avoid injuring tooth buds or the base of the skull. The constructed nasal passage was

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extended to the nasopharynx. After the new nasal airway was drilled, a modified high Le Fort I maxillary osteotomy was performed to advance the midface anteroinferiorly. Then the inferior segment was advanced 3 mm forward and 2.5 mm downward. This widened the newly developed nasal airway and midface advancement. The skin flap partially covered only the anteroinferior inlet of the newly developed nasal passage, and the raw surfaces on the roof and the sides of the newly constructed nasal passage were left. Although the new nose was temporarily functional after surgery, the absence of respiratory mucoepithelium tissue could lead to stenosis of the nasal passage and concomitant impairments in nostril breathing.

After nasal construction surgery for arhinia patients, a commercial nasal balloon stent (Rapid Rhino; ArthroCare ENT, Austin, TX) was inserted into the nose for hemostasis of the nasal passage. However, the commercial nostril stent models used in pediatric patients were unsuitable for this purpose, because these stents are only designed for the nostrils, and not for the nasal cavity.

**Design, Fabrication, and Implantation of a Patient-Specific Nasal Stent**

To create a nasal silicone stent customized for the arhinia patient, we used three-dimensional (3D) printing technology, which was less time consuming than traditional methods. The stent was designed in conjunction with the patient’s postoperative computed tomography (CT) image analysis (slice thickness = 0.6 mm). The stent included an oval-shaped surface fitted to the size and shape of the nasal passage (from the nostrils to the nasal cavity), with an inner offset surface for constructing a windpipe. The pipeline was connected from the nasal cavity to the right nostril part, with an angle of approximately 100° between the longitudinal axes. The customized silicone stent was created using an indirect-3D (i3D) printing process, which consisted of printing a 3D mold, injecting the desired material into the mold cavity, and removing the mold (Fig. 2). The stent mold, designed using Boolean operators, included both inlets and outlets for the injection of silicone resin, and was divided into eight parts to allow for easy removal following hardening of the resin. Computer-aided drawing models of the mold parts were converted into a stereolithography file format, and subsequently imported into the 3D-printing system (Dimension SST; Stratasys, Eden Prairie, MN). ABSplus (Dimension SST; Stratasys) was used for the 3D printing.
used as the underlying material for the printed mold components. The silicone resin (Q7-4840; Dow Corning, Midland, MI) was injected into the inlet of the assembled mold. Then the resin was solidified at 160 °C in a preheated oven for 10 minutes, and maintained at room temperature. Finally, the mold components were removed to obtain the stent (Fig. 3A). The overall time required for fabrication of the stent was about 7 days.

Three weeks after implantation of the commercial nasal balloon stent, we implanted the customized, patient-specific i3D-printed silicone nasal stent (from the nostrils to the nasal cavity) under general anesthesia during the third operation, to maintain the newly developed nasal airway. The stent was placed into the new nose, with a tagging string pulled out of the mouth to allow full insertion of the i3D-printed nasal stent. As illustrated in the CT images (Fig. 3B), the stent was mounted stably for 2 months after implantation, with the shape and size of the nasal passage largely maintained. About 2 months after implantation, the stent was removed using a straight clamp during outpatient care. The newly generated nasal passage was then measured using an endoscope (Olympus, Tokyo, Japan) following stent removal. The nasal airway to the nasopharynx and the mucus-covered respiratory mucosa were seen during endoscopy (Fig. 4).

DISCUSSION

Implant products are widely used in medicine, such as prosthesis, and they are often purchased from commercial manufacturers. Despite such high demand, the supply of surgical implants is outweighed by the demand, because each product needed by the individuals varies from each other depending on their physical symptoms.

The advantages of 3D-printing technology—including an easy and rapid processing ability relative to subtractive manufacturing—allow for its wide range of use in fields such as industry, architecture, and military. When applied in the treatment of deformities, 3D-printing technology can be an innovation; precisely shaped implants and scaffolds printed in a short time can reduce the total duration of the operation. The simplified operational procedures can also provide both the patients and the surgeons with higher satisfaction. That is, 3D-printing technology would be one of the best methods for fabrication of implants for the treatment of deformities.

Arhinia, as we mentioned above, is one of the rarest deformities, which has been reported only in 30 cases by the 21st century. Use of stents in the patient’s reconstructed nose is necessary for preventing stenosis, but designing the stent is quite complicated, and thus commercialized implants are not suitable.

Here, we designed a customized nasal stent for an arhinia patient based on CT images. Using a molding process with 3D-printing technology, the stent was constructed from silicone. Biomedical-grade silicone resin provided by Dow Corning may be used for direct printing of an unsophisticated structure. However, constructing the stent from directly printed silicone resin is a labor-intensive and time-consuming process, because printing of a stent, a bent tubular body, requires temporary supporting parts, and the resin is difficult to dispense continuously from a nozzle due to its high viscosity. Thus, the stent was devised using i3D-printed injection molds, which can be produced in small batches given the viscosity and complexity of the structure.

We facilitated the prevention of restenosis in the new nasal passage by promoting a functional mucoepithelium and physically suppressing regeneration of granulation tissue. The fabricated stent was implanted for 2 months, and after stent removal, newly regenerated mucoepithelium tissue from the nasopharynx was detected in the constructed nasal passage, and respiratory function remained normal.

Our patient-specific i3D-printed nasal stent facilitated the attenuation of stenosis in the constructed nasal airway passage, through regeneration of mucoepithelium

![Fig. 3. (A) The designed stent model and the printed stent. (B) Computed tomography images of the newly regenerated nasal passage with a patient-specific nasal stent. Stent insertion is depicted within the yellow box.](image1)

![Fig. 4. Endoscopic view of the regenerated nasal respiratory mucosa after removal of the stent.](image2)
tissue over a period of approximately 2 months. Following stent removal, nasal airway function and diameter (6 mm) were maintained for an additional 8 months and 3 years, as shown in the endoscopic view (Fig. 5).

CONCLUSION

A customized nasal stent was constructed for an arhinia patient using our i3D-printing technology. The stent was implanted into the patient's nose for 2 months to prevent stenosis of the newly developed nasal passage and facilitate regeneration of respiratory mucoperiosteal tissue in the constructed nasal passage. This study illustrates the potential effectiveness of customized nasal stents for arhinia patients using 3D-printing technology. To evaluate the long-term efficacy of nasal reconstruction, we will periodically assess the function, shape, and size of the patient's constructed nasal passage. Patient-specific i3D-printed nasal stents would represent a new standard of care for the management of arhinia. Here, we described the first case of i3D-printed nasal stent implantation in an arhinia patient, with potential applicability worldwide to a variety of specific clinical cases requiring custom-made medical devices. Moreover, in the future, if this technology could also be applied to external nasal structure reconstruction, it would be possible to provide the best functional and aesthetic result for the patient.

BIBLIOGRAPHY