How I Do It

Management of Midnasal Stenosis With Infant Surgically Assisted Rapid Palatal Expansion (iSARPE)

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INTRODUCTION

Neonates are obligate nasal breathers; therefore, stenosis at any level can be associated with significant respiratory distress. Drill-out techniques are described for the management of pyriform aperture stenosis and choanal atresia; however, midnasal stenosis is a common associated feature not addressed by these procedures. Failed surgical management of pyriform stenosis and choanal atresia may be due to scarring or unrecognized concomitant midnasal stenosis. Additionally, craniofacial disorders such as Apert and Crouzon syndromes can be associated with nasal stenosis without pyriform aperture or choanal stenosis.1,2

Surgically assisted rapid palatal advancement (SARPE) is a technique of palatal distraction that has been used to provide space for crowded maxillary dentition and in maxillary hypoplasia.3 More recent usage includes adult obstructive sleep apnea (OSA) patients.4 Rapid maxillary expansion without osteotomies has been described in pediatric patients with OSA.5,6 SARPE has not been described in early childhood.

We describe the use of infant SARPE (iSARPE) to expand the nasal cavities in two patients with different pathologies causing midnasal stenosis. This technique has not previously been described for nasal stenosis, and our early results have been promising for their efficacy.

METHODS

Planning

Preoperative low-dose maxillofacial computed tomography (CT) scans were obtained via fine cuts in the axial plane with sagittal and coronal reconstructions. Imaging confirmed stenosis extending to the midnasal passage. Three-dimensional reconstructions were used to plan the osteotomies (Fig. 1a) and screw placement.

A palatal dental impression was used to create a custom acrylic palatal expansion device fitting the contour of the high arched palate. The device was designed and fabricated by a dedicated craniofacial team orthodontist to fit into the palate with an 11-mm expansion screw (Leone, Florence, Italy) at the center of the device. After review of imaging, screws were planned to orient from medial on the device angle obliquely to engage the maxilla between the lateral nasal wall and tooth buds. Holes were drilled and then tapped with self-tapping screws (DePuy Synthes, Zuchwil, Switzerland); screw length varied based on device, gingiva, and bone thickness, with superior and oblique orientation.

SURGICAL TECHNIQUE

The nasal cavity was examined with a zero-degree 2-mm endoscope, and 1% lidocaine plus 1:200,000 epinephrine was injected in the superior gingivolabial sulcus. The anterior maxillary spine, pyriform aperture, face of maxilla, and infraorbital foramina were exposed. Dissection continued into the nasal cavity, elevating the nasal floor and inferior turbinate mucosa. If stenotic, the bony pyriform aperture was widened with the skeeter drill. Urethral sounds were inserted intranasally, and lateral pressure was applied to widen the midnasal stenosis via turbinate out-fracture. Improved airway caliber was confirmed before expansion by reduced resistance to these sounds and the ability to introduce sounds of a larger diameter.

Bilateral osteotomies were planned, starting above the head of the inferior turbinate and carried laterally just below the infraorbital nerve foramina, through the face of the maxilla anterior to the rudimentary maxillary sinus, and to the edge of the maxilla. A midline osteotomy was planned through the superior surface of the hard palate behind the alveolar ridge, between the vomer and the palatal shelves. Planned osteotomies were marked on the maxilla and performed with the Piezoelectric saw (DePuy Synthes) after confirming position of toothbuds on preoperative imaging. The pterygoids were not fractured. The cartilaginous septum was dissected off the anterior maxillary spine to allow the midline osteotomy. A finger was placed on the oral hard palate mucosa to palpate the blade of the saw, tenting...
the mucosa to confirm complete cut without mucosal violation. The 2- and 4-mm straight osteotomes were used to complete the midline osteotomy and open the suture line between the vomer and the palatal shelves.

Two-layer closure of the incision was performed (periosteum and gingiva). A custom palatal expansion device was placed, and preplanned self-tapping screws were used to secure the device (Fig. 1b). The device was opened two turns to engage the device and begin the process of expansion.

Active distraction commenced on postoperative day 2 with the device manually supported, with two turns (0.5 mm) performed twice daily until resistance was encountered, and then (0.25 mm) twice daily. Patients were each expanded 10 to 11 mm. The device was placed, and preplanned self-tapping screws (periosteum and gingiva). A custom palate expansion device was placed, and preplanned self-tapping screws were used to secure the device (Fig. 1b). The device was opened two turns to engage the device and begin the process of expansion.

RESULTS

Case 1

TF, a term female infant, presented at birth with respiratory distress requiring intubation. The nasal cavity would not admit a 10 French catheter. A narrow pyriform aperture, absent labial frenulum, and high-arched palate with midline ridge were noted on physical exam. Otolaryngology was consulted, and CT/magnetic resonance imaging imaging was obtained. The patient was diagnosed with pyriform aperture stenosis and solitary median maxillary central incisor syndrome. The patient’s pyriform aperture measured 3.5 mm at its narrowest, and 8.0 mm from lateral nasal wall to lateral nasal wall half-way between choana and pyriform aperture.

The patient was extubated and managed medically with steroid nasal drops; however, the patient continued to have episodes of cyclical respiratory distress and cyanosis and noisy breathing with feeding. Surgical management was therefore discussed, and the patient underwent pyriform aperture drill-out in addition to iSARPE to manage the midnasal stenosis.

Postoperatively, the patient was distracted as described above. Nasal stents (Nose-Fit, Moscow, Russia) were placed to help with soft tissue collapse at the internal nasal valve. The patient had no further cyanotic episodes and did not require oxygen postoperatively. The patient required one dose of narcotic and was otherwise managed with acetaminophen for pain control. The patient remained in the hospital until the parents were comfortable performing distraction. By the first follow-up visit, the patient had been distracted 10 mm, and the larger second custom stents would not remain in place due to the increased size of the nasal cavity. Endoscopic images are shown in Figure 2.

Case 2

LR, a term female infant, was transferred to our tertiary pediatric facility at 1 day of life for respiratory distress and multiple congenital anomalies. Computed tomography imaging showed stenotic but patent choanae (11.9 mm), relative pyriform aperture (8.7 mm), and significant midnasal stenosis, with the patient’s midnasal vault measuring 11.7 mm from the lateral nasal wall to the lateral nasal wall halfway between choanae and pyriform aperture (Fig. 3a). The patient was diagnosed with Apert syndrome. Initially, the patient was managed medically with nasal stents and steroid drops; however, the patient’s nasal congestion and feeding continued to worsen. iSARPE was therefore offered to the patient’s family for management of the multilevel nasal stenosis, in addition to pyriform aperture drill-out and nasal balloon dilation. This was performed at 10.5 weeks of age.

The patient remained in hospital for 4 days postoperatively to improve parental comfort with distraction. Postoperatively, the patient underwent distraction as detailed above. The patient developed yellow nasal crusting suspicious for infection at 1 month postoperatively, which was treated with ointment and discontinuation of nasal stents. A postoperative CT performed for the patient’s cranial vault showed improvement in the nasal airway (Fig. 3b). Postoperative polysomnography showed an obstructive apnea hypopnea index (OAH) of 2.7 (mild sleep apnea).

Results remained stable, with one additional balloon dilation performed during the general anesthetic for...
craniosynostosis repair at 25 weeks of age. The patient unfortunately passed away at home at 32.5 weeks of age due to a witnessed aspiration event at 6.5 weeks after the cranial vault procedure.

DISCUSSION

The traditional approach to congenital nasal pyriform aperture stenosis is a sublabial incision with drill-out of the overgrown maxilla. Dilation with cervical dilators has also been described as a successful intervention, but follow-up to date has been short and re-dilations frequently appear to be required.7 Choanal atresia may be managed with posterior septectomy with or without drilling of the bony plate. These patients may require revision procedures either due to scarring or unaddressed midnasal stenosis. Computed tomography evidence of narrowing of the nasal cavity distinct from the pyriform aperture is often seen in patients with pyriform aperture stenosis. This midnasal vault narrowing in pyriform aperture stenosis patients has been illustrated in a previous study of nasal cavity dimensions by Reeves et al. LW-1 (lateral nasal wall measurement 1) in this study was described as the measurement from the lateral nasal wall to lateral nasal wall measured in the axial plane halfway between the choana and pyriform aperture.8 Mean LW-1 measurement in pyriform aperture stenosis patients in this study was 8.7 ± 0.2 mm, as compared to 13.5 ± 0.28 mm in controls.8 This measurement technique was chosen for our patients as well due to its reliability and ease. Using these landmarks, patient 1’s midnasal vault measured 8.0 mm preoperatively, and patient 2’s was 11.7 mm.

SARPE has been used for several indications in orthognathic surgery in adults. Subjective improvement in nasal symptoms has been incidentally noted with this procedure. A 2015 study of patients undergoing SARPE showed at 6 months that their Nasal Obstruction Symptom Evaluation scores were either stable or improved.9 When rapid maxillary expansion without surgery is used in pediatric patients, an increase in nasal dimensions is also noted by acoustic rhinometry.10 This procedure has never been described with nasal stenosis as the primary indication for surgery. iSARPE also differs in that pterygoid fracture is not performed. The pterygoid plates may act as a fulcrum for anterior expansion, but more research is needed to elucidate the effect of this procedure on the nasal cavity.

Apert syndrome may be a condition in which iSARPE for nasal obstruction will be useful, with many

Fig. 2. Preoperative (a) and 3-month postoperative (b) endoscopic images. The middle turbinate could not be visualized prior to the procedure and was freely visible bilaterally postoperatively.

Fig. 3. Preoperative (a) and 3-month postoperative (b) midnasal dimensions in patient 2, measured at the pyriform (*) and halfway between pyriform aperture and choana #. Measurement * increased from 8.7 mm preoperatively to 14.4 mm postoperatively, and measurement # increased from 11.7 mm preoperatively to 15.4 mm postoperatively.
of these patients presenting with choanal, pyriform, and midnasal stenosis. A previous Apert series showed that 60% of the patients required nasal airway interventions, many requiring multiple revision surgeries.

Pyriform aperture stenosis repair also often requires revision surgery. A systematic review found 14% of patients required revision for scarring versus unrecognized multi-level narrowing. In acute situations, failed nasal stenosis repairs can necessitate tracheostomy. iSARPE has the potential to address multiple levels of nasal stenosis concurrently. Longer-term data is needed to examine whether revision is required, although to date our patients have had stable results.

We did not observe significant complications in our iSARPE patients; however, this procedure is not without risk: a retrospective cohort study revealed nearly half of adult patients following SARPE experienced complications from postoperative pain to dental anomalies. Although we cannot yet comment on the long-term dental outcomes in our patients, we did not observe any of the significant perioperative complications described, such as pain and hemorrhage. No readmissions or reoperations have been required. Device malfunction is a potential risk that must be carefully mitigated. Rotational force during active distraction could dislodge the device. We found the device stability increased as distraction progressed.

Our series is very small, with only two patients having undergone this procedure to date. However, early postoperative results are promising. We were unable to follow one patient long-term due to mortality unrelated to a nasal procedure. Future directions include lengthening the duration of follow-up, refining the indications for this procedure, identifying ideal timing of device removal, and examining outcomes in more patients.

CONCLUSION

iSARPE is a newly described technique for the management of midnasal stenosis in infants. Early results are promising; however, a larger series is needed.

BIBLIOGRAPHY