Trans-nasal puncture for bilateral congenital choanal atresia

Alireza Alam Sahebpor, Vajihe Ghaffari, Ali Reza Elisay
Department of Pediatric Surgery, Mazandaran University of Medical Science, Sari, Iran

Correspondence: Dr. Alireza Alam Sahebpor, Department of Pediatric Surgery, Booali Sina Hospital, P.O. Box: 4715838477, Pasdaran Boulevard, Sari, Iran, E-mail: alireza_alam@yahoo.com

ABSTRACT

Aims: There are many surgical techniques to repair bilateral congenital choanal atresia, among them trans-nasal approach with simple puncture, dilation and stenting technique is most frequently used. We describe a series of trans-nasal puncture and stenting technique, by using pediatric urethral sounds and nalaton N 14.

Materials and Methods: We used this technique in 10 patients with equal sex distribution, between 1997 and 2004 in our hospital. Mean age at operation was 3.5 days. Mean duration of stenting was 33.6 days (2-8 weeks) and patients were followed for an average duration of 28.8 months after surgery.

Results: Three patients (30%) developed re-stenosis after stent removal, which was easily managed by simple dilatation and re-stenting for more two weeks. There was a minimal operative bleeding with no infection or central nervous system trauma.

Conclusion: This safe, simple operation by using pediatric urethral sounds is a non-expensive way to treat this anomaly with a low complication rate.

KEY WORDS: Choanal atresia, neonate, stents

Congenital choanal atresia (CCA) is a relatively rare anomaly, occurring approximately one in 5000 to 8000 live births. It occurs in females twice as often as males and unilateral CCA is twice as common as bilateral CCA.

Bilateral CCA presents as a medical emergency at birth. Obvious airway obstruction, stridor and cyclic cyanosis with crying are the usual presentations and a temporizing measure such as oral airway, McGovern nipple or intubation is required prior to definitive surgical treatment. There are several surgical approaches available for its correction; among them trans-nasal approach is favored by most surgeons. Perforation of the atretic plate is done by trocars, chisels, drills and lasers with or without assistance of endoscopes and because of the frequent recurrence of the stenosis; the use of nasal stenting has became usual.

Here, we describe a trans-nasal puncture and stenting technique, using pediatric urethral sounds, without endoscope, in 10 neonates with bilateral CCA and its results.

MATERIALS AND METHODS

From 1997 to 2004, we had 10 neonates with equal sex distribution with bilateral CCA in Booali Sina hospital in Sari, Iran. All neonates presented with severe respiratory distress, cyclic cyanosis with crying and stridor.

Complete physical examination was performed and the diagnosis was confirmed by inability to pass a small stent through the nares. We did not do pre-operative computed tomography (CT) scan on our patients. They were managed initially by placing a large-hole nipple or an airway to keep the mouth open. No patient needed orotracheal intubation or tracheostomy.

Surgical technique

Surgery was performed under general anesthesia and orotracheal tube was placed. At the time of surgery, mean age was 3.5 days. Pediatric urethral sounds were used to perforate the obstructing plate. They have the ideal curvature to follow the sloping contour of the nasal floor [Figure 1].

We started with smallest sound, which is normally N 4. The sound is passed along the floor of the nose, staying against the septum to avoid penetrating the basal sphenoid. Placing the fingers of the other hand into the mouth assessed the direction of the sound. When the
obstructing plate was touched, the sound was carefully forced through it with uniform pressure. The procedure was repeated for the other side. Then the perforations were gradually dilated with larger sounds and both sides were dilated serially to prevent shifting of the septum. After dilatation, nalaton N 14 was passed over the smallest sound through the perforated membrane and pushed enough forward to allow a 1 cm protrusion of the nalaton into the nasopharynx.

The distal part of the sound was brought out of nares and cut, so about 2 cm of the sound was out of nares. This projection of the stent did not interfere with breast-feeding and also there was no case of nasal regurgitation.

In the first five cases, the stent was secured to each ala with 3-0 silk. Because of difficulty in maintaining the tubes, in the other five patients we sutured the proximal parts of the nalatons to each other with silk suture that they would remain in the nasopharynx. In this way, we did not need to suture them to ala and hence, there was no chance for the stents to be accidentally dislodged and causing trauma to the ala.

Post-operative care
After operation intra-venous cephallexin was given for 2 days and nasal stents were irrigated with 1cc normal saline and suctioned with syringe every 6 h to keep them patent. The patients were discharged 3 days post-operation. Before discharging from hospital, the parents were educated on appropriate care of the stents. They were taught to irrigate and suction the stents regularly with a syringe.

After 4 weeks, in the operating room and under general anesthesia the stents were brought out and new stents were inserted. We taught the parents to remove the stents for half an hour in the morning and also in the evening for another 2-4 weeks. In this period, beta-mathasone nasal drop was given and used by parents instilling one drop in each nasal cavity once a day for 2 weeks. We did not use mitomycin C in our patients. After final removal of the stents, we followed our patients for an average of 28.8 months (ranging from 8 to 52 months) to see whether they would develop complication or not. In our study records were analyzed with respect to: (1) Sex, (2) age at the time of surgery, (3) intra-operative and post-operative complications, (4) duration of stenting, (5) recurrence, (6) duration of follow-up [Table 1].

RESULTS
Our patients were five girls and five boys, all with bilateral CCA who were managed initially by placing a large-hole nipple or an airway to keep the mouth open. They were operated between day 2 and 7 of life. Pediatric urethral sounds were used to puncture the plate and nalaton N14 was placed as a stent in all patients. Duration of stenting was between 2 and 8 weeks with a mean of 33.6 days.

There were no intra-operative complications such as significant nasal hemorrhage, septal perforation, skull base damage and cerebrospinal fluid (CSF) leak or midbrain trauma.

Table 1: Details of our patients’ data

<table>
<thead>
<tr>
<th>Year</th>
<th>Sex</th>
<th>Age at surgery (days)</th>
<th>Stenting period (weeks)</th>
<th>Follow-up period (months)</th>
<th>Complication</th>
</tr>
</thead>
<tbody>
<tr>
<td>1997</td>
<td>M</td>
<td>6</td>
<td>6</td>
<td>36</td>
<td>Left side re-stenosis in the first month of follow up</td>
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<td>1997</td>
<td>F</td>
<td>7</td>
<td>2</td>
<td>8</td>
<td>Right side re-stenosis in the first month of follow up</td>
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<tr>
<td>1998</td>
<td>M</td>
<td>2</td>
<td>3</td>
<td>52</td>
<td>Right side re-stenosis in the second month of follow up</td>
</tr>
<tr>
<td>1998</td>
<td>M</td>
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<td>8</td>
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<td>1999</td>
<td>F</td>
<td>2</td>
<td>5</td>
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<td>2001</td>
<td>M</td>
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<td>2002</td>
<td>F</td>
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<td>2003</td>
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<td>2004</td>
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<td>2004</td>
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anesthesia and new stents were placed and parents were taught to remove these stents two times a day. Patients were followed for an average of 28.8 months (ranging from 8 to 52 months) and in this period, three patients (30%), two of them in the first month of follow up and the other one in the second month of follow up, developed unilateral complete obstruction based on symptoms of nasal obstruction and inability to pass a stent [Table 1]. They were all among our first five patients. They were re-stented for more 2 weeks; and all responded well to re-stenting and dilatation.

**DISCUSSION**

Congenital choanal atresia is a relatively rare anomaly; it may present as a part of a non-random association of congenital malformations grouped together as CHARGE syndrome (cloboma, heart disease, choanal atresia, developmental delays, genital hypoplasia and ear anomalies) with a reported incidence of 1:10,000 birth.[9] There is a controversy about the etiology of CCA. Persisting bucconasal or bucopharyngeal membrane are possible causes.[4] And reports have indicated that 90% of CCAs are bony, while 10% are membranous, but recent reviews using CT studies suggest that most atresias are mixed and all membranous atresias have some bony component.[5-7]

CCA was first described by Roederer in 1755[8] and the first documented operation for CCA was performed by Emmerth in 1851. He used the trans-nasal puncture by a curved trocar to perforate the membrane.[6,10]

As newborns are obligatory nasal breathers for the first few weeks of life, bilateral CCA is a medical emergency requiring immediate attention at birth and some measures including oral airway, McGovern nipple or intubation should be done as soon as possible to temporally relieve obstruction and this should be followed by a definite surgery. In our series, all patients were initially managed by large-hole nipple or oral airway and there was no need for intubation before operation. Concerning clinical symptoms, CCA can be diagnosed in several ways; the simplest way is failure to pass a catheter through the nose into the nasopharynx. Diagnosis of CCA can be confirmed by choanogram and/or CT scan. CT scan is the radiolology of choice for the diagnosis of CCA. It can show nature, thickness and the extent of the atretic plate.[2] As mentioned, we did not perform choanogram or CT in our cases, but according to the symptoms, complete physical examination and failure to pass the small catheter we excluded conditions like severe choanal stenosis and reached the diagnosis of CCA.

Up to now four different surgical approaches have been described for surgical treatment of CCA: (1) trans-nasal; (2) trans-palatal; (3) trans-septal; and (4) trans-antral.[11,12] The two most popular techniques that are used in newborn infants with CCA are trans-palatal and trans-nasal approaches. The trans-palatal approach offers a wider filed of vision, but can cause maldevelopment of the upper dental arch with a crossbite deformity and in a study, dental malocclusion (crossbite) was reported in 52% of patients, younger than 5 years, who were managed this way and this approach has been reserved for revision cases in older patients; and recently it has been largely dispensed with in favor of endo-nasal approaches.[3]

Other options include the trans-nasal puncture dilation, stenting by using mastoid curretts, trocares, Farea dilators and CO2 and KTP lasers to puncture the plate and trans-nasal endoscopic technique.[13,14]

Endoscopic endonasal surgery for CCA was first reported by Stankiewicz,[4] followed by EL-Guindy et al,[15] Kamel[16] and Cumberworth et al.[7] This technique allows excellent visualization. A 2.7 mm endoscope is used in the neonates and using the endoscope minimizes the recurrence rate and the need for revision surgery. However, the most experienced endoscopist will be challenged by the narrow nasal cavity of the neonates and further by septal deviation, turbinate hypertrophy and a high arched palate.[17]

In a recent survey of American Society of Pediatric Otolaryngologist (ASOP) members, 85% of respondents utilized endoscopic technique, 60% trans-palatal and 17% puncture technique for bilateral CCA that shows the controversy that exist on the procedure of choice.[17]

The ideal procedure should be safe, quick and simple with minimal blood loss and high rate of success.[2] In our experience, we performed trans-nasal puncture stenting technique by using curved urethral sounds without endoscope. Because of the soft nature of the bone in neonates, there was no difficulty in perforating the atretic plate and gradual dilatation was done in the same setting with larger urethral sounds. In this technique, the operation is simple and the operation time is short, approximately 15-20 min; also we do not need sophisticated equipments and operation is done easily, with very infrequent complications.

In most reports some kind of stent is used to prevent re-stenosis post-operatively. Mostly 3.0 or 3.5 mm endotracheal tube is used as a stent; other materials used for this purpose are, soft silicone, self inflating silastic tube, silicon suction tube and metal reinforced rubber silicone tubes.[15,18-20] We used nalaton N 14 bilaterally in
our cases and we did not have any difficulty with them. The infants were comfortable and tolerated them well.

Usually there is problem in fixing the stents in a way that there would be no trauma to ala in addition to preventing them from dislodgment and pushing out. In our first five cases, we sutured the stent to the ala, but it caused some trauma to it in three patients. In addition, we could not keep the stent in place because they were accidentally dislodged. And may be this is the reason for re-stenosis in these three patients. Therefore, we changed our stent fixing technique; in this modification we sutured the distal ends of natalons, which were in nasophrynx with silk suture together and by doing this, the stents were fixed in place and there was no chance of accidental dislodgment and consequently the chance of re-stenosis diminished.

Although the use of nasal stenting is accepted by most surgeons, the duration of stenting is variable ranging from 4 to 6 weeks in most reports and up to 6 months in some publications.[16,18] In a retrospective study, reported by Gujrathi et al, there were 52 patients with bilateral CCA; a simple puncture, dilatation and stenting were performed by using a modified endotracheal tube with a diameter of 3 or 3.5 mm for a mean stenting time of 12 weeks; only three patients required re-stenting because of persistent unilateral stenosis, so they concluded that long period of stenting is essential for long-term success. They also noted that careful use of trans-nasal puncture and dilatation with stenting in the treatment of neonates with bilateral CCA yields success rates in achieving long-term nasal patency comparable to trans-nasal endoscopic techniques.[16] Sadek et al used the trans-nasal puncture technique in eight neonates with bilateral CCA. The duration of stenting in this series was 6-8 weeks and in his follow-ups, there was only one patient who needed dilatation.[24] Unlike Gujrathi et al,[24] we could not establish the necessity of long period stenting for long term success; and we suppose, it should be investigated in future studies with more patients.

Moreover, despite the use of certain stents, the development of granulation tissue in the choana is a frequent occurrence, which causes re-stenosis. And that is why there is no uniformity of opinion as to the efficacy of one type of stent or method of surgery above another. The use of mitomycin (an anti-proliferative and anti-tumor aminoglycoside), which is used intra-operatively to prevent scar tissue and granulation formation post-surgery has still to be established as beneficial.[25] In a study by Kubba et al they had 46 patients who underwent the trans-nasal puncture technique by using endoscope through the mouth and stenting with Portex endotracheal tube for an average stenting duration of 3 months. They used mitomycin C nasal drop after stent removal in some patients; there was no difference in regard of re-stenosis in the group with mitomycin compared with dose with out mitomycin.[14] In our experience we did not use mitomycin and instead, we used betamethasone nasal drop for 2 weeks.

Furthermore, the main complications of the puncture technique are bleeding, infection and central nervous system (CNS) trauma (including CSF leakage and meningitis from perforation and fracture of the perpendicular plate of the ethmoid cribiform plate). In the series reported by Gujrathi et al there was no CNS complication and no incidence of significant nasal hemorrhage or infection.[25] In our experience, although, there was minor bleeding intra-operatively that resolved after placing the stents, we did not have any significant complication. Careful puncture with uniform pressure and using the curvature of pediatric urethral sounds and staying against the septum and ethmoid palate are possible reason for the low complication rate.

CONCLUSION

Bilateral CCA is a medical emergency that needs immediate stabilization and very soon definitive surgery. The trans-nasal puncture and stenting that we performed by using pediatric urethral sounds seems to be a safe, quick and simple technique that can be used in the first week of life, with minimal complications. We think that with this simple operation, by using instruments, available in every pediatric surgery center, would be a good substitute for sophisticated and expensive operations that use telescopes and micro-drills, which may not be available in developing countries.

REFERENCES

Sahebpor AA, et al.: Trans-nasal puncture for bilateral congenital choanal atresia


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