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Congenital midnasal stenosis: Conservative management

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ABSTRACT

Introduction: Congenital midnasal stenosis (MNS) is an extremely rare disease which may be life threatening, and shows difficulty in diagnosis and management. This case series summarizes superiority of using intranasal mometasone furoate spray (IMS) and continuous positive airway pressure (CPAP) to treat nasal obstruction in neonates with MNS.

Methods: This study reviewed six consecutive cases of MNS.

Results: Three patients were treated with IMS and CPAP. Two patients were treated with endoscopic balloon dilatation without stenting, followed by IMS and CPAP due to persisting nasal obstruction after the operation. One patient was treated with endoscopic balloon dilatation without stenting alone.

Conclusion: This study is the first to review the use of CPAP and IMS as an effective therapy for some patients with MNS. It suggests that conservative management with CPAP and IMS may be an alternative therapeutic option to surgery.

1. Introduction

As neonates are obligate nasal breathers, any pathology restricting the normal airflow may be a life threatening condition and requires urgent management. Persistent nasal obstruction in newborns presents at birth despite having a normal tracheobronchial tree. The manifestations include failure-to-thrive and intermittent respiratory distress worsening during feeding followed by relief with crying.

The source of the neonatal nasal obstruction can vary from congenital nasal masses to bony stenosis or mucosal congestion. Congenital bony stenosis can be classified according to the side of the stenosis as anterior stenosis or congenital pyriform apertura stenosis (CPAS), middle or midnasal stenosis, posterior stenosis or choanal atresia [1].

Midnasal stenosis (MNS) is a rare clinical entity due to bilateral overgrowth midway through the intranasal cavity, resulting from excessive infolding of the nasal septum [2]. There have been few cases of midnasal stenosis and treatment reported in literature.

Before surgical treatment for midnasal stenosis, conservative management methods such as the use of nasal drops, a McGovern pacifier or nasal airway can be preferred. Surgical management should be reserved for infants with persistent apnea, failure to-thrive, and in cases of resistance to conservative therapies. Patel et al. mentioned successful conservative management in thirteen infants with an average time-to-resolution of 126 days [3]. However, endoscopic assisted lateralization of the inferior turbinate and nasal stenting under general anesthesia has been described to successfully recover patency of the nasal airway [1].

The purpose of this study is to describe the clinical presentation, radiologic diagnosis, and management of six newborn infants with this rare pathology.

2. Material and methods

This is a retrospective review of all patients presenting with congenital MNS, diagnosed and referred to our tertiary care hospital between 2014 and 2019. Cases were analyzed and reviewed for details including patient demographics, type of coexisting anomalies, clinical presentation, initial management, computed tomography (CT) findings and outcomes.

The Ethics Committee of Umraniye Training and Research Hospital approved the study, and informed consent was taken from the parents.

MNS was diagnosed when these infants had clinical features of airway obstruction such as difficult breathing, failure to thrive, nasal congestion, alternating episodes of cyanosis which improved whenever they cried, with resistance or impossibility encountered during passing a nasogastric tube or a fibroscope in either nostril. The diagnosis was confirmed with a craniofacial CT-scan which revealed a bony narrowing on both sides about midway inside the nasal cavities as well as the absence of choanal atresia (Fig. 1). There were a total of 6 patients treated (2 girls; 4 boys). Two of them (case 3 and 5) had been diagnosed with laryngomalacia. The other 4 patients did not have any comorbid

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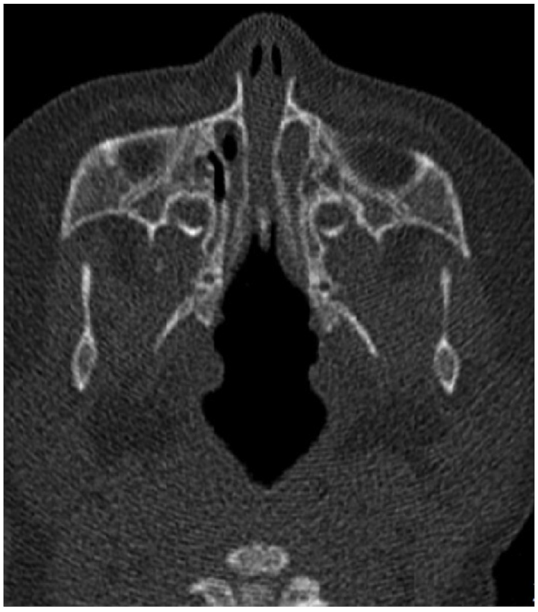

Fig. 1. Axial CT scan of patient 4 demonstrating midnasal stenosis.

diseases. All patients had respiratory symptoms at birth. Cases 1, 3, 4, 5, 6 had documented severe respiratory distress with apnea/cyanosis. Clinical features are as summarized in Table 1.

One patient (case 3) was intubated immediately after birth due to severe respiratory distress. Case 3 and 5 underwent supraglottoplasty and endoscopic widening of the nasal cavity with balloon dilatation without stenting. Another patient (case 4) underwent endoscopic widening of nasal cavity with balloon dilatation without stenting as an initial management. Other patients were initially treated with nasal decongestants; (%0,025) oxymetazoline hydrochloride, (once daily for 3 days), saline 0, 9% nasal drops (twice daily), intranasal mometasone furoate spray (IMS) (once daily) and CPAP (continuous positive airway pressure). Pediatric endocrinologists were consulted for observation or management of side effects of mometasone nasal spray. The duration of IMS and CPAP treatment was modified according to symptoms.

2.1. Surgical procedure

Surgical correction was performed under general anesthesia via an endoscopic endonasal approach. Direct laryngoscopy and tracheoscopy were performed to exclude the presence of a possible additional airway disease. Following application nasal decongestion with diluted (1/100.000) epinephrine, a 2,7 mm 0° nasal endoscope (Storz Corporation, Germany) was used during the operation. Midnasal stenosis was enlarged using a 5 mm balloon sinoplasty catheter (Relieva Solo Sinus Balloon Cathater™) for both nasal passages. The balloon was inflated by saline up to 80 mm H2O pressure for 2 min. No stent was inserted into the nasal cavities. Additionally case 3 and 5 underwent supraglottoplasty. Patients were followed by administration of saline and xylometazoline hydrochloride nasal spray (0,05%) for three days in the postoperative period.

3. Results

A total of 6 patients were diagnosed with MNS. 4 patients were male and 2 patients were female (Table 1).

All 6 patients had persistent feeding difficulties, failure to thrive and respiratory symptoms including at least one of a variety of symptoms (stridor, noisy breathing, chest retractions, desaturations, and tachypnea). All patients were symptomatic at birth, one patient (case 3) had required intubation due to stridor, four patients (case 1, 4, 5, 6) had

Table 1
Patients' characteristics (ND = nasal decongestant; CPAP: continuous positive airway pressure SI = 0.9% saline irrigation, IMS: intranasal mometasone furoate spray).

Case	Sex	Presentation	Immediate management	Duration of IMS treatment	Comorbid disease	CT scan findings	Age at diagnosis	Follow up age outcome
1	Female	Severe respiratory distress, cyanosis	ND, SI, CPAP (30 days), IMS	45 days	None	Bony narrowing of the anterior and mid nasal region	30 days	36 months asymptomatic
2	Male	Moderate respiratory distress, no cyanosis	ND, SI, NCPAP (20 days), IMS	30 days	None	Isolated bony narrowing of the mid nasal region	25 days	30 months asymptomatic
3	Male	Severe respiratory distress, failure to thrive, cyanosis	İntubated, supraglottoplasty, nasal cavity balloon dilatation, anti-reflux, ND, SI, CPAP (20 days), IMS	45 days	Laringomalacia	Bony narrowing of the anterior and midnasal region	15 days	22 months asymptomatic with peg
4	Female	Severe respiratory distress and cyanosis	Nasal cavity balloon dilatation, ND, SI, IMS	45 days	None	Isolated bony narrowing of the mid nasal region	20 days	9 months, mild OSAS
5	Male	Severe respiratory distress, failure to thrive, cyanosis	Supraglottoplasty, nasal cavity balloon dilatation, Anti-reflux, ND, SI	None	Pulmonary atresia	Isolated bony narrowing of the mid nasal region	6 months	19 months, asymptomatic
6	Male	Severe respiratory distress and cyanosis,	ND, SI, CPAP (20 days), IMS.	45 days	None	Isolated bony narrowing of the mid nasal region	20 days	9 months asymptomatic

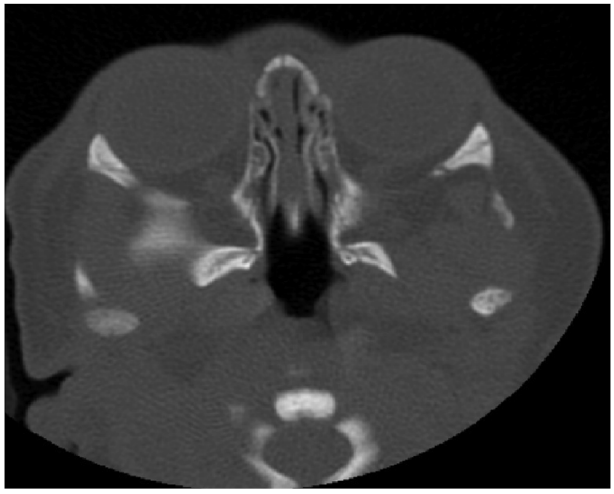

Fig. 2. CT scan of patient 6 showing midnasal stenosis.

severe and one patient (case 2) had moderate respiratory symptoms.

Flexible endoscopy (with 2, 2 mm scope) was attempted but was not successful due to the narrowness of each nasal passage. But 5F nasogastric tube was able to be passed into both nasal passages. Therefore, paranasal CT scan was performed to confirm the diagnosis of MNS. On paranasal tomography images, noticeable narrowing was observed in the airway at the level of the inferior turbinate in the central part of the nasal cavity (Fig. 2).

Patient 1 was a term female infant. Age at diagnosis of MNS was 30 days. Initially, she was managed medically with nasal oxymetazoline hydrochloride (once daily, 3 days), saline 0,9% nasal drops (twice daily, 3 months), IMS (once daily, 45 days) and nCPAP (30 days). The patient's nasal obstruction and feeding improved at 60 days of age (Fig. 3).

Patient 2 was a male term infant. He had moderate respiratory distress and no synosis at birth. Age at diagnosis of MNS was 25 days. Oxymetazoline hydrochloride nasal spray (0,05%) (once daily, 3 days), IMS (once daily, 30 days), saline 0,9% nasal drops (twice daily, 45 days) and CPAP (20 days) were used initially for treatment. The symptoms improved at 50 days of age.

Patient 3 was term male infant and had respiratory distress requiring intubation at the first day of life. He was diagnosed with midnasal stenosis and laryngomalacia at 15 days of age. Supraglottoplasty and endoscopic widening of the nasal cavity with balloon dilatation without stenting were performed at 17 days of age. The patient was extubated at three days after surgery and used CPAP for 20 days. Nasal obstruction persisted after the operation. Saline 0,9% nasal drops (45 days, twice daily) and xylometazoline hydrochloride nasal spray (0,05%) (3 days, once daily) and IMS (once daily, 45 days), were used. The symptoms improved at two months of age.

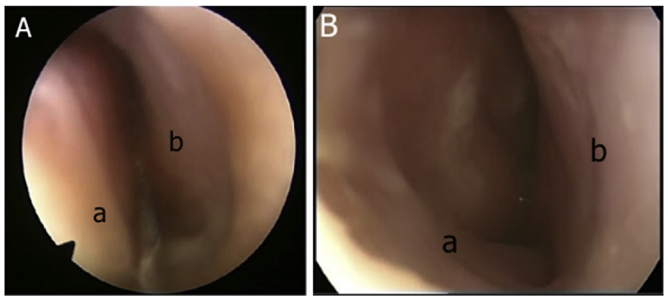

Fig. 3. (Patient 1) Endoscopic images of right nasal cavity A: Before conservative treatment, B: Two months after conservative treatment. a: inferior turbinate, b: nasal septum.

Patient 4 was a term female that presented with stridor, tachypnea, and retraction of the chest occurred immediately after birth. She was diagnosed with MNS at 20 days of his age. Endoscopic widening of the nasal cavity with balloon dilatation without stenting was performed two days after MNS was diagnosed. She was extubated immediately after the operation. The symptoms improved moderately, but on 45 days of life the symptoms returned due to synechiae between inferior turbinate and septum in her left nasal passage. After using oxymetazoline hydrochloride nasal spray (0,05%) (3 days, once daily), IMS (once daily, 30 days), saline 0,9% nasal drops (45 days, twice daily), the symptoms improved mildly again. On 90th day of her life she weaned completely. Now she is nine months old and has synechiae in her left nasal passage. We plan a surgical correction after the age of three.

Patient 5 was a male born at 35 weeks gestation. He was kept in the NICU (neonatal intensive care unit) and had cardiac surgery due to pulmonary atresia. An otorhinolaryngological examination was requested at 6 months of age because of stridor, difficulty in feeding and recurrent apnea. Nasal examination and CT scan revealed midnasal stenosis. Under general anesthesia, we performed endoscopic widening of the nasal cavity with balloon dilatation without stenting and supraglottoplasty. He was extubated immediately after the operation. Saline 0,9% nasal drops (20 days, twice daily) and xylometazoline hydrochloride nasal spray (0,05%) (3 days, once daily) were used. All symptoms resolved 10 days after surgery (Fig. 4).

Patient 6 was a male one-month-old child. He was referred to our department with inspiratory stridor with subcostal, intercostal retractions and poor feeding. Nasal examination and CT scan demonstrated midnasal stenosis. He was initially treated with nasal oxymetazoline hydrochloride nasal spray (0,05%) (3 days, once daily), IMS (once daily, 45 days), saline 0,9% nasal drops (45 days, twice daily) followed by CPAP for 20 days. His symptoms improved at 2 months of age.

4. Discussion

Nasal airway obstruction is a life-threatening condition. Neonatal

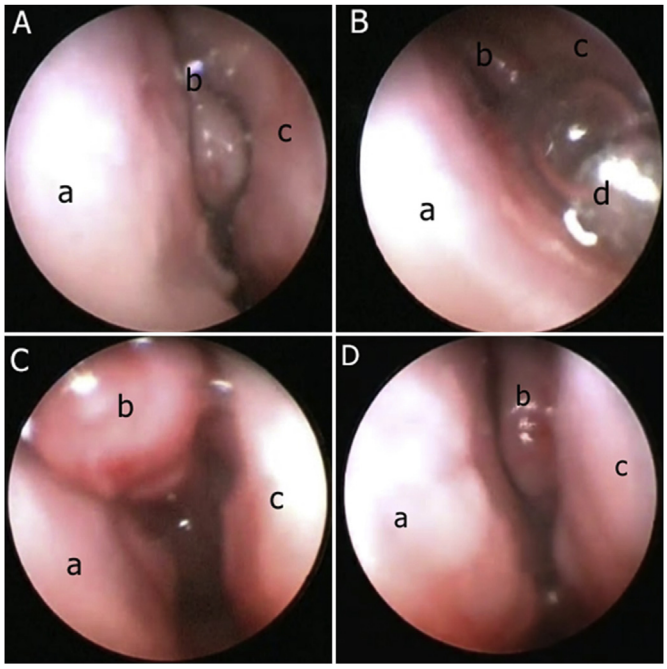

Fig. 4. (Patient 5) Endoscopic images of left nasal cavity A: Intraoperative view before balloon dilatation, B: Intraoperative view with balloon dilatation C: Intraoperative view post balloon dilatation D: Postoperative view 2 months later. a: nasal septum, b: middle turbinate, c: inferior turbinate, d: balloon catheter.

rhinitis, viral upper respiratory tract infections and possibly milk/soy allergies are among the most common causes of neonatal nasal obstruction. Less common reasons include congenital bony nasal anomalies such as choanal atresia and stenosis, nasal agenesis, pyriform apertura stenosis, midnasal stenosis, traumatic deformation, and septal deviation. Other causes for neonatal nasal obstruction are neoplasms [4].

Congenital midnasal stenosis is an uncommon clinical entity characterized by narrowing of the fibrous or bone tissue in the nasal cavity between priform apertura and choana. It is secondary to unequal growth of the lateral wall of the nose or excessive infolding of the nasal septum [1]. In the exclusion of choanal atresia and priform apertura stenosis, care must be taken that midnasal stenosis is not overlooked in patients with nasal obstruction.

Congenital bony nasal obstructions are diagnosed clinically by difficulty to pass a size 5 French Gauge catheter. Fiberoptic nasal endoscopy provides important data regarding the site of the obstruction but gives minimal information regarding the extent, severity and type of stenosis. Bony stenosis at the nasal aperture blocking out the view of inferior turbinate is a frequent finding of congenital priform aperture stenosis (CPAS). If the fiberoptic scope can be passed past the anterior end of the inferior turbinate for a certain distance proximal to stenosis, but the middle turbinate cannot be visualized despite the absence of any gross septal deviation, midnasal stenosis is to be considered [5]. CT scanning is essential to confirm the diagnosis. Normal nasal dimensions for neonates and infants based on CT findings have recently been published [6]. However there is no standardized measurement for midnasal stenosis [1]. CT will confirm either isolated bony narrowing of the midnasal region or arrowing associated with stenosis of the remainder of the nasal cavity [7].

For midnasal stenosis conservative approach with frequent nasal saline irrigation and suctioning until 6 months of age at which point the midface has grown enough to relieve the obstruction is recommended. A McGovern pacifier and oral airway can be used to assist with feeding. Surgical treatment may be considered in patients with repeated intubation, failure to extubation and conservative treatment [8]. Use of CPAP and IMS may offer an effective alternative to surgery. This study is the first to suggest CPAP and IMS to be used as an alternative therapeutic option to surgery for some patients with MNS. Conservative treatment with CPAP and IMS was successful in five of our patients.

Specific studies addressing the effect of topical application of steroids consistently demonstrate a preservation of the normal histologic appearance of nasal mucosa. Therefore the use of intranasal steroids in the neonatal period is relatively safe [9]. The mechanism of improving nasal obstruction with intranasal corticosteroid (ICS) is uncertain. The anti-inflammatory properties of ICS may suggest a significant underlying inflammation of the nasal mucosa in patients with MNS. In addition, mucosal atrophy from steroidal properties of ICS could play a role in the improvement [10].

Nasal directional positive airway pressure using the Infant Flow System (Viasys) provides directional flow continuous positive airway pressure and is used as an alternative to intubation and ventilation in neonates [11]. Its system uses directional flow continuous positive airway pressure which reduces the work of breathing by decreasing airway fluctuation due to Coanda effect [12]. CPAP is typically used to treat respiratory distress syndrome, apnea of prematurity and respiratory failure after extubation. Moreover, CPAP is useful for upper airway obstruction [13]. In our case series, CPAP via nasal mask was effective for relieving nasal obstruction due to bilateral midnasal stenosis [14].

In our series the conservative treatment with IMS and CPAP was successful in three of our patients without surgery. Their breathing improved within three weeks. No complication was observed due to CPAP and IMS. The other 3 patients were managed with balloon dilatation without stenting. In patient 4, iatrogenic nasal adhesions appeared. In patient 3, nasal obstruction persisted after the surgery. His symptoms improved with CPAP and IMS. Ozdemir et al. reported a case of bilateral MNS, in which balloon dilatation without stent was done. They repeated the procedure because of the recurrence of symptoms without any complications [15]. In the literature, there are numerous case reports about the success of balloon dilatation with stenting in children with congenital nasal stenosis [7,15,and16]]. Balloon dilatation with stenting may be an alternative surgical treatment for the failure of conservative management due to causing lateral displacement of the inferior turbinate. But we do not suggest balloon dilatation without stenting due to the recurrence of symptoms and synechiae.

Some of the potential side effects of using ICSs are adrenal insufficiency, immunosuppression and metabolic changes which may be minimized by close monitoring by endocrinologists, by using the lowest possible dosage that provides the effective therapeutic outcome by early and slow tapering. A case series was reported by Collins et al., in 2013, in which one of their patients had iatrogenic Cushing's disease and adrenal suppression due to intranasal dexamethasone treatment [10]. We used mometasone furoate which has a lower bioavailability than dexamethasone. No side effects were observed in our patients. Further experience with using intranasal steroids in infants will help elucidate its optimal dose in a treatment for MNS.

The limitations of this study were small number of subject and the lack of control subjects. Proper knowledge and experience with using IMS at this age is mandatory with regards the efficacy and safety of IMS.

5. Conclusions

In infants with nasal obstruction related respiratory distress who do not have choanal atresia and pyriform aperture stenosis, midnasal stenosis should be considered. Intranasal mometasone furoate spray and CPAP may be a safe, successful and effective procedure as an initial therapy for MNS. Further experience with infants will help elucidate the optimal dose of ICS for treatment.

Declaration of competing interest

No potential conflict of interests relevant to this paper was reported.

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