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Letter

Congenital Maxillomandibular Synechiae: Anesthetic Approach with Ketamine and Dexmedetomidine Sedation for Surgical Division of Synechiae and Tracheostomy

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Dear Editor,

Congenital maxillomandibular syngnathia of the jaws, is a rare condition that has a wide variety of presentations (1). As the fusion may be unilateral or bilateral, in some cases, bony or soft tissue adhesions between the mandible and maxilla may be accompanied by the cleft lip and palate, aglossi and popliteal pterygium.

Restricted mouth opening can lead to problems with feeding, swallowing and respiratory functions therefore affecting the growth of the infant. Early diagnosis and management is important to reduce these problems and avoid the risk of aspiration. Hereby, we present a case with congenital maxillomandibular synechiae, with buccal mucosa and the gingival adhesions who underwent surgical division and successful tracheostomy procedure under ketamine and dexmedetomidine sedation.

A 1-day-old female newborn (weight 3180 g) suffering from lack of mouth opening since birth was brought to our plastic surgery clinic for evaluation. The patient was referred to the Plastic and Reconstructive surgery department on account of inability to open the mouth and difficulty in feeding secondary to her anatomical defect. The mother had an uncomplicated pregnancy and received proper prenatal care. In family's medical history, it was learned that their first child had also same congenital anomaly and died 3 months after birth due to respiratory complication.

In examination, it was seen that the lips could be separated but the maxillary and the mandibular gingivae were fused completely at the occlusal level anteriorly and the baby's mouth opening was approximately 1 mm.

She had micrognathia and in addition, bony and mus-

cular involvement in the synechiae was noted in radiological evaluation. Cranial and abdominal ultrasonography in newborn was normal and patent ductus arteriosus was detected in echocardiographic evaluation. Laboratory blood exam values were within normal limits.

It was decided to divide synechiae surgically. At the same time surgical preperations were made for the possibility of tracheostomy. The patient was monitored with noninvasive blood pressure, five-channel EKG and pulse oxymeter in the operating room. After pretreatment with atropine 0.02 mg.kg⁻¹ i.v. ketamine 1 mg.kg⁻¹ was administered. Dexmedetomidine (Precedex, Abbott) was started as infusion at $0.7 \cdot 1 \mu g.kg^{-1}.h^{-1}$. After sedation with dexmedetomidine and ketamine the patient had spontaeous breathing and was oxygenated (4 L.min⁻¹) via a flexible nasopharyngeal tube.

Following local anesthetic injection (1% lidocaine with 1:100,000 epinephrine) into the adhesive areas, the synechiae were divided using surgical scissors. The mouth opening was immediately improved to approximately 7 mm, however, there was still a restriction in mouth movement. In addition, there was also a cleft hard palate. Therefore, it was decided to perform gastrostomy with tracheostomy in order to protect the patient from aspiration of regurgitated gastric contents and feeding material.

Tracheostomy was performed with dexmedetomidine sedation while spontaneous breathing was maintained. Effective sedation was provided during this procedure and well tolarated by the patient. A minimal change in vital signs during the surgery and serious adverse events (such as bronchospasm, desaturation, bradycardia) were not observed. After succesfull tracheostomy, the dexmedetomi-

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dine infusion was discontinued and anesthesia was maintained with sevoflurane 1.2% -1.5% and 50%:50% oxygen and air. Operation lasted 45 minutes. At the end of the surgery, patient was transferred to the pediatric intensive care unit without any respiratory adverse effect.

Congenital alveolar synechiae is an uncommon defect. It can be found in any part of oral cavity and generally includes fibrous and supportive tissue such as muscle or bone and membranes or bands of epithelium (2). It can be associated with microglossia, micrognathia, cleft palate or syndromes like Vander de Woude, and Pierre-Robin (1, 2).

Fibrous alveolar synechiae without bone or muscle abnormalities have good outcomes when properly excised. But, this defect is becoming more complex for surgical intervention in cases involving bone tissue as in our patient. In order to treat the patient effectively, it is very important to secure the airways and ensure adequate nutrition until enough mouth opening is provided. Therefore, we had decided for gastrostomy and tracheostomy in our patient.

Dexmedetomidine is a more selective α 2-adrenergic receptor agonist than clonidine. It has been reported to provide reliable rapid and stable sedation, analgesia, and hemodynamic modulation, without clinically significant respiratory depression (3). Hypotension and bradycardia are the most prominent and often dose-limiting side effects of dexmedetomidine.

Ketamine is another popular anesthetic which has both sedative and analgesic properties. But ketamine, can also induce adverse cardiostimulatory effects and postoperative delirium when used as a sole sedative agent (4).

There are various publications in the literature regarding the use of ketamine and dexmedetomidine for sedation in different pediatric surgical procedures (3). Bozdogan et al. (5) showed that bolus doses of ketamine 1 mg.kg⁻¹ and dexmedetomidine 1 μ g.kg⁻¹ followed by a continuous infusion of dexmedetomidine 0.7-1 μ g.kg⁻¹.h⁻¹ provided excellent anesthetic conditions to perform caudal anesthesia and inguinal hernia repair in three high-risk infants.

Levanen and colleagues (4) also emphasized that the combined use of ketamine and dexmedetomidine will neutralize the expected side effects of each other.

The sympathomimetic properties of ketamine helped to maintain hemodynamics in our patient and prevent bradycardia and hypotension. Also, in our cases, there were no episodes of airway obstruction reported, the respiratory rate remained stable.

Especially in cases where mask ventilation and possibility of intubation is difficult, maintenance of spontaneous breathing and airway is important. The characteristic of dexmedetomidine and ketamine to provide sedation with minimal respiratory depression makes them suitable anesthetic agents for this procedure. In sumary, the combination of dexmedetomidine, ketamine and local anesthesia provided an adequate depth of anesthesia and analgesia for surgical division and tracheostomy operation in our patient. We believe that the combined use of dexmedetomidine and ketamine should be reminded by different case presentations and clinical trials in order to achieve the best clinical scenario for possible benefits and safety profile.

References

- Cerrati EW, Ahmed OH, Rickert SM. Isolated congenital maxillomandibular synechiae. *Am J Otolaryngol.* 2015;36(5):707–9. doi: 10.1016/j.amjoto.2015.04.008. [PubMed: 25957707].
- Gartlan MG, Davies J, Smith RJ. Congenital oral synechiae. Ann Otol Rhinol Laryngol. 1993;102(3 Pt 1):186–97. doi: 10.1177/000348949310200305. [PubMed: 8457120].
- Tobias JD. Dexmedetomidine and ketamine: an effective alternative for procedural sedation? *Pediatr Crit Care Med.* 2012;13(4):423–7. doi: 10.1097/PCC.0b013e318238b81c. [PubMed: 22067985].
- Levanen J, Makela ML, Scheinin H. Dexmedetomidine premedication attenuates ketamine-induced cardiostimulatory effects and postanesthetic delirium. *Anesthesiology*. 1995;82(5):1117–25. doi: 10.1097/00000542-199505000-00005. [PubMed: 7741286].
- Bozdogan N, Sener M, Caliskan E, Kocum A, Aribogan A. A combination of ketamine and dexmedetomidine sedation with caudal anesthesia during incarcerated inguinal hernia repair in three highrisk infants. *Paediatr Anaesth.* 2008;18(10):1009-11. doi: 10.1111/j.1460-9592.2008.02566.x. [PubMed: 18811854].