

At 23 months of age, the patient underwent an MRI of the face with and without contrast which showed a right anterior sublingual prominence (Fig. 1B). No inflammatory changes were appreciated. At 2 years of age, the patient experiences self-limited fluctuating floor of the mouth swelling with associated sialorrhea.

Case 3

Patient 3 underwent MDO at 7 weeks of age for postnatally diagnosed RS associated with cleft palate and 22q11 duplication (Supplemental Table 1, Supplemental Digital Content 1, <http://links.lww.com/SCS/F262>). His internal distractor devices were removed without perioperative complications. His caregiver first endorsed sublingual swelling 1 year following MDO. On physical examination, a left soft, nontender, anterior 1.5 cm sublingual lesion with moderate mass effect on the tongue was appreciated. The caregiver denied infectious symptoms. An MRI of the face was added to a previously scheduled noncontrast MRI brain. Imaging showed left sublingual prominence without associated inflammatory changes (Fig. 1C). At 20 months, the patient underwent palate repair. The patient continues to experience persistent sialorrhea that accompanies the sublingual swelling which is managed conservatively.

DISCUSSION

Potential postoperative complications of MDO are well documented in the literature and include infection, facial nerve injury, and tooth injury.^{2,4-6} Recurrent episodes of the floor of mouth swelling following MDO, as observed in this case series, is a rare complication. The patients' surgeries were performed by 3 different surgeons across 2 unique institutions, making it unlikely that the injuries were due to a specific technical or mechanical error. The timeline for the presentation was delayed and varied for each patient, presenting 2 to 12 months following removal of the distraction device and before palatoplasty. No patients were reported to have tenderness, pain, fevers, or coinciding dental work associated with the lesions, making an infectious etiology unlikely. The immediate postoperative period was uneventful except for one patient who experienced a unilateral hardware detachment that was addressed early without sequela.

In evaluating the possible etiology, we considered sublingual salivary duct dilation. The submandibular or sublingual glands or ductal system could be interrupted during the active distraction phase. It is possible that the entity we describe herein is an intermittent sublingual gland or ductal obstruction leading to edema due to prior trauma from surgery or the distraction phase. Morovic et al⁷ described 2 cases of sublingual gland hypertrophy following MDO in early 2022. These patients struggled with oral intake and cleft palate repair due to tongue displacement and underwent surgical excision to address the mass effect.⁷ As our patients' lesions have yet to be biopsied or excised we cannot reliably confirm gland hypertrophy or fibrosis. In addition, in contrast, our patients' floor-of-mouth swelling has fluctuated in presence and laterality, unlike Morovic et al's⁷ description.

Other diagnoses considered include ranula, sublingual sialocele, and lymphoepithelial cyst. Although imaging was not consistent with ranulas as they appear as well circumscribed lesions.^{8,9} In addition, while the back-to-back surgical opening of Risdon incisions and dissection to place and subsequently remove internal distractors in a neonate could traumatically hinder normal floor of mouth lymphatic

drainage, lymphoepithelial cysts are unlikely to fluctuate or cause sialorrhea.¹⁰

Presently, all 3 children continue to experience intermittent sublingual edema. Despite these symptoms, the patients are feeding and growing appropriately, therefore surgical intervention has been deferred and the patients are managed conservatively. If our patient's lesions are akin to Morovic et al's description,⁷ we aim to present conservative management as an alternative to surgical excision in MDO postoperative sublingual gland hypertrophy.

CONCLUSION

These 3 cases represent a rare sequela following neonatal MDO. The chronicity and intermittent nature of symptom onset reinforces the need for continued follow-up even in cases without immediate postoperative complications. All cases are presently being monitored conservatively without indication of surgical intervention. Future studies should be directed toward capturing the incidence of the intermittent floor of mouth swelling or sublingual gland hypertrophy in this population and further characterizing the long-term outcomes and causes of this phenomenon.

REFERENCES

1. Hsieh ST, Woo AS. Pierre Robin sequence. *Clin Plast Surg* 2019;46:249–259
2. Morrison KA, Collares MV, Flores RL. Robin sequence: neonatal mandibular distraction. *Clin Plast Surg* 2021;48:363–373
3. Khansa I, Hall C, Madhoun LL, et al. Airway and feeding outcomes of mandibular distraction, tongue-lip adhesion, and conservative management in Pierre Robin sequence: a prospective study. *Plast Reconstr Surg* 2017;139:975e–983e
4. Master DL, Hanson PR, Gosain AK. Complications of mandibular distraction osteogenesis. *J Craniofac Surg* 2010;21:1565–1570
5. Paes EC, Mink van der Molen AB, Muradin MS, et al. A systematic review on the outcome of mandibular distraction osteogenesis in infants suffering Robin sequence. *Clin Oral Investig* 2013;17:1807–1820
6. Lam DJ, Tabangin ME, Shikary TA, et al. Outcomes of mandibular distraction osteogenesis in the treatment of severe micrognathia. *JAMA Otolaryngol Head Neck Surg* 2014;140:338–345
7. Morovic CG, Torres J, Jorquera C. Unusual mandibular distraction complication in two neonatal Pierre Robin sequence patients. *J Craniofac Surg* 2022;33:e201–e203
8. Edwards RM, Chapman T, Horn DL, et al. Imaging of pediatric floor of mouth lesions. *Pediatr Radiol* 2013;43:523–535
9. Bowers EMR, Schaitkin B. Management of mucoceles, sialoceles, and ranulas. *Otolaryngol Clin North Am* 2021;54:543–551
10. Yang X, Ow A, Zhang CP, et al. Clinical analysis of 120 cases of intraoral lymphoepithelial cyst. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2012;113:448–452

Surgical Management of Infection Secondary to Cemento-osseous Dysplasia

Farin Ebrahimi, DDS,* Faraz Ebrahimi, DDS,† and Jingang An, DDS, MD*

Abstract: Cemento-osseous dysplasia (COD) of the jaws generally has no clinical manifestations when asymptomatic, thus requiring no treatment. However, secondary infection in COD requires surgical intervention. This study was focused on the evaluation of the surgical treatment of COD patients with secondary infections. The clinical data of COD patients with secondary infections, treated at the Peking University, Hospital of Stomatology between March 2021 and June 2022, were retrospectively reviewed. The data included age, sex, lesion characteristics, number of surgeries, and surgical outcomes. Seven COD patients with secondary infections underwent curettage, and the wounds were repaired using local soft tissue flaps, such as the buccal fat pad. Four of the patients had primary wound healing, while 3 presented with wound dehiscence. Healing occurred in 2 of these 3 patients after ~1 month of dressings. The remaining patient showed no improvement after 9 weeks, and underwent a second surgery, which led to primary wound healing. In conclusion, secondary infection in COD is an indication for surgical intervention, which may arrest the disease progression.

Key Words: Cemento-osseous dysplasia, secondary infection, surgical management

Cemento-osseous dysplasia (COD) is a benign fibro-osseous lesion that originates from the periodontal ligament and causes the normal bone to be replaced by a cementum-like structure, followed by the calcification of bone and cementum. In accordance with the location as well as the number of lesions, COD is divided into 3 types. A COD lesion found at the apical area of the anterior teeth is referred to as periapical COD. Focal COD appears as a solitary lesion, commonly in the posterior region, but can occur in any part of the maxilla or mandible. Florid COD involves bilaterally symmetric lesions in multiple jaw quadrants.¹

It has been reported that COD is more common among middle-aged Black females, but also occurs in Asians and Caucasians.² A systematic review by MacDonald-Jankowski included 158 florid COD patients, showed that only a small

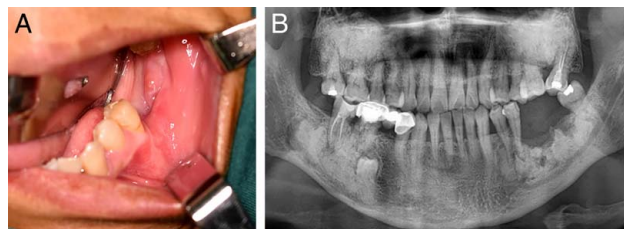


FIGURE 1. (A) A 55-year-old female experienced repeated swelling, pain, and pus discharge for 10 months from nonhealing left lower first molar extraction socket. (B) Preoperative panoramic radiograph showing florid cemento-osseous dysplasia in the maxilla and mandible, along with alveolar bone resorption and destruction in the edentulous area of the posterior left mandible.

percentage were male, more than half were Black, and one-third were Asian.³

Cemento-osseous dysplasia generally shows no clinical manifestations. However, it might be discovered during dental checkups when a radiologic examination is required. Asymptomatic COD requires regular follow-up, with surgical intervention only when symptoms begin to appear. Cemento-osseous dysplasia with a secondary infection may cause swelling, pain, pus discharge, fistulas, and sequestrum formation, and requires surgical intervention.

The present study reviewed the clinical characteristics and surgical outcomes of COD patients with secondary infections.

METHODS

This study retrospectively analyzed the demographic data, clinical data which included age, sex, lesion characteristics, number of surgeries, surgical outcomes, COD characteristics, and postoperative state of the COD patients who underwent surgery at the Peking University, Hospital of Stomatology between March 2021 and June 2022.

The inclusion criteria were clinical and imaging manifestations consistent with the diagnosis of COD, and typical features of secondary infection. After surgical treatment, the pathologic diagnosis was confirmed, and the clinical data were completed. Preoperative informed consent was obtained from all patients.

The patients received surgery under general anesthesia. The lesion was fully exposed intraorally, involved teeth were extracted, infected bone was completely removed, and healthy bone around the lesion was preserved as much as possible. A local soft tissue flap, such as the buccal fat pad, was used to fill the bone defect, and the wound was tightly sutured. Postoperative panoramic radiographs and spiral computed tomography were used to evaluate bone healing. The patients were instructed to come back for follow-ups between the different

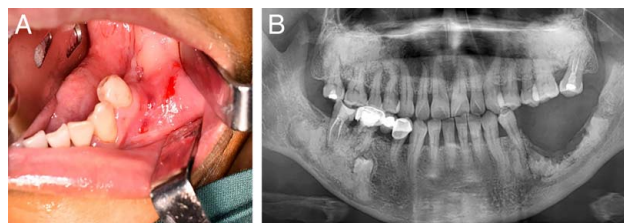


FIGURE 2. (A) After the first surgery, the same patient shown in Figure 1 presented with intraoral fistula and pus discharge. Wound dressing was done 1 to 2 times a week with no improvement. (B) A panoramic radiograph 9 weeks after the first operation showed irregular bone resorption in the operated area (left posterior mandible).

From the *Department of Oral and Maxillofacial Surgery, Peking University School and Hospital of Stomatology South Avenue Zhongguancun, Haidian District, Beijing, PR China; and †Department of Prosthodontics, Peking University School and Hospital of Stomatology, Beijing China.

Received May 22, 2023.

Accepted for publication June 1, 2023.

Address correspondence and reprint requests to Jingang An, DDS, MD, Department of Oral and Maxillofacial Surgery, Peking University School and Hospital of Stomatology, NO. 22 Zhongguancun Nandajie, Haidian District, Beijing 100081, PR China; E-mail: anjingang@126.com

The authors report no conflicts of interest.

Supplemental Digital Content is available for this article. Direct URL citations are provided in the HTML and PDF versions of this article on the journal's website, www.jcraniofacialsurgery.com.

Copyright © 2023 by Mutaz B. Habal, MD

ISSN: 1049-2275

DOI: 10.1097/SCS.00000000000009556

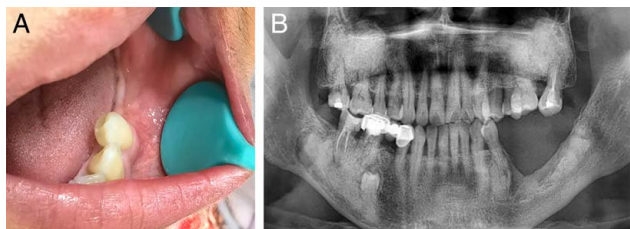


FIGURE 3. (A) The same patient 15 months after the second surgery showed good intraoral wound healing. (B) Panoramic radiograph 15 months after the second surgery showing smooth bone surface in the operated area.

time periods of 1 week, 1 month, and 3 months. During postoperative follow-ups, bone healing was assessed by panoramic radiographs and spiral computed tomography.

RESULTS

A total of 7 COD patients with secondary infections underwent surgical treatment, including 5 females and 2 males, with a mean age of 66.5 (range: 49–84) years. Six of the patients (85.7%) were diagnosed with florid COD, while 1 (14.3%) was diagnosed with focal COD (Fig. 1). Delayed extraction wound healing occurred preoperatively in 5 patients, 1 patient developed a local infection because of a removable denture, and 1 patient had residual roots accompanied by pus discharge.

The surgical method involved resection, buccal fat pad transfer to repair the defect, and tension-free suturing to close the wound. The median postoperative hospital stay was 3 days. Primary intraoral wound healing occurred in 4 patients. The remaining 3 patients showed signs of wound infection and dehiscence from the first of follow-ups postoperatively; the wounds received treatment by irrigation using normal saline and packed with iodoform gauze. In 2 cases, the symptoms significantly improved after 3 weeks of treatment, and the wounds healed completely after 6 weeks. The remaining patient had poor treatment results, with persistent fistula and suppuration (Fig. 2). This patient underwent a second surgery after 9 weeks, which led to successful primary wound healing. Over 6 to 24 months of follow-up, successful healing of the intraoral wounds occurred in all cases, with no limitation of the mouth opening (Fig. 3). The patient characteristics are listed in supplemental Table 1 (Supplemental Digital Content 1, <http://links.lww.com/SCS/F269>).

DISCUSSION

Cemento-osseous dysplasia points to a group of benign fibrous bone lesions, in which either tooth-bearing or edentulous alveolar bone gradually matures by calcification.^{4,5} In asymptomatic COD cases; the affected teeth are immobile, the pulp is vital, and the surrounding gingiva appears normal.⁶ It is often found incidentally on dental imaging.

As cementum-like calcifications steadily deposit during the maturation of COD lesions, secondary infection risks also rise. Literature suggests that infections are more likely after trauma that causes bone exposure, such as tooth extraction, close to the lesion.^{7,8} The symptoms of secondary infection can appear in the resorbed edentulous alveolar bone because of high bone density and reduced blood circulation. Trauma may also be caused by dentures, masticatory force imbalance, and inappropriate treatment can also cause progressive alveolar bone atrophy, bone exposure, and secondary infection.⁹ Odontogenic infection involves COD around the root of the tooth. The immune response of the COD bone is weakened due to osteosclerosis, which leads to secondary infections.¹⁰

Cemento-osseous dysplasia without secondary infection generally has no obvious clinical manifestations, and surgical treatment is mainly required when the lesion develops symptoms or infection. It has been reported that osteosclerotic lesions exposed to the oral flora are more prone to infections.¹¹ Invasive treatment for infected COD can be applied in the appearance of pain, suppuration, and osteolysis regardless of bone sequestration.

The most common treatment of secondary infection requires surgical resection of the diseased cortical bone, removal of poorly vascularized infected lesions, restoration of local bone blood supply, and a prescription for antibiotics.^{7,8,12}

Reduced vascularization and increased bone density in COD make the affected area prone to infections and necrosis, which becomes a challenging treatment for surgeons. In this study, approximately half of the secondary infection patients had poor postoperative wound healing, and required further treatment. During surgery, the infected bone should be completely removed, and the incision should be tightly sutured. A local soft tissue flap may be used to fill the bone defect and assist in wound closure.¹³ A buccal fat pad was used in the present study to repair the wound in 5 patients. In cases such as the ones presented in this study, patients that presented poor wound healing, routine irrigation, and iodoform gauze changes during follow-ups did help to promote healing. However, if the wounds fail to show improvement after about 3 to 4 months of dressing changes, secondary surgery can be considered.

Clinically, COD with secondary infection is different from primary suppurative osteomyelitis, which is caused by highly virulent bacteria in cases with reduced immunity. In suppurative osteomyelitis, enucleation of the sequestra and debridement generally leads to normal wound healing because the surrounding bone is normal.

Asymptomatic COD found incidentally on imaging does not require any treatment except regular follow-up and observation. Good oral hygiene can reduce the risk of endodontic and periodontal infections, and thus the risk of odontogenic infections. It is also necessary to avoid trauma, such as that caused by surgery or dentures, to prevent bone exposure and subsequent infection.⁹

It has been reported that COD with secondary infection is more common in patients aged >40 years.^{4,14} This indicates that an increase in dental problems with aging increases the susceptibility to odontogenic infections. To control the occurrence or re-occurrence of infection secondary to COD, clinicians should explain the importance of oral hygiene, apprise patients of regular checkups, and inform COD patients of the consequences of secondary infections.

REFERENCES

1. Waldron CA. Fibro-osseous lesions of the jaws. *J Oral Maxillofac Surg* 1993;51:828–835
2. MacDonald-Jankowski DS. Florid cemento-osseous dysplasia: a systematic review. *Dentomaxillofac Radiol* 2003;32:141–149
3. Huh JK, Shin SJ. Misdiagnosis of florid cemento-osseous dysplasia leading to unnecessary root canal treatment: a case report. *Restor Dent Endod* 2013;38:160–166
4. Pereira DL, Pires FR, Lopes MA, et al. Clinical, demographic, and radiographic analysis of 82 patients affected by florid osseous dysplasia: an international collaborative study. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2016;122:250–257
5. El-Naggar AK, Chan JKC, Takata T, et al. The fourth edition of the head and neck World Health Organization blue book: editors' perspectives. *Hum Pathol* 2017;66:10–12
6. Min CK, Koh KJ, Kim KA. Recurrent symptomatic cemento-osseous dysplasia: a case report. *Imaging Sci Dent* 2018;48:131–137

7. Manoharan G, Manoharan GV. Florid osseous dysplasia-report of two cases and review of literature. *J Clin Diagn Res* 2017;11:ZD21–ZD24
8. Macdonald-Jankowski DS. Focal cemento-osseous dysplasia: a systematic review. *Dentomaxillofac Radiol* 2008;37:350–360
9. Fenerty S, Shaw W, Verma R, et al. Florid cemento-osseous dysplasia: review of an uncommon fibro-osseous lesion of the jaw with important clinical implications. *Skeletal Radiol* 2017;46:581–590
10. Carulli C, Innocenti M, Brandi ML. Bone vascularization in normal and disease conditions. *Front Endocrinol (Lausanne)* 2013;4:106
11. Brannon RB, Fowler CB. Benign fibro-osseous lesions: a review of current concepts. *Adv Anat Pathol* 2001;8:126–143
12. Alsufyani NA, Lam EW. Osseous (cemento-osseous) dysplasia of the jaws: clinical and radiographic analysis. *J Can Dent Assoc* 2011;77:b70
13. Salehi-Nik N, Rezai Rad M, Kheiri L, et al. Buccal fat pad as a potential source of stem cells for bone regeneration: a literature review. *Stem Cells Int* 2017;2017:8354640
14. Consolaro A, Paschoal SRB, Ponce JB, et al. Florid cemento-osseous dysplasia: a contraindication to orthodontic treatment in compromised areas. *Dental Press J Orthod* 2018;23:26–34

A Simple and Minimal Invasive Method in Reduction of Depressed Nasal Bone Fracture by Using a Foley Catheter Ballooning Technique

Ke Xue, MD, PhD,*† Kai Liu, MD, PhD,* and
Lin Qi, MD, PhD*

Abstract: The nasal bone fracture is the most common type of facial bone fracture. Closed reduction with metal reduction instrument is commonly conducted for the treatment of a type II nasal bone fracture. The authors defined a new catheter dilation technique and used it in patients with type II depressed nasal bone fractures. Preoperative and postoperative

nasal appearance and radiologic examination of the patients were compared. There was a statistically significant improvement in the nasal appearance of all patients. No recurrence or dorsal irregularity has been observed. This new, easily applicable catheter dilation method of closed reduction may be a simple and less invasive solution to treat type II nasal bone fractures.

Key Words: Catheter dilation, closed reduction, facial bone fracture, Foley catheters, nasal bone fracture

The nasal bone fracture account for half of the facial bone fracture, which is the most common type of facial bone fracture.¹ To treat nasal bone fracture properly, it is important to diagnose and evaluate the nasal bone fracture. The nasal bone fracture was classified into 4 types according to Fred,² and open reduction and closed reduction are 2 classic therapeutic strategies to treat nasal bone fracture. Closed reduction is commonly conducted for the treatment of a nasal bone fracture unless it is accompanied by a severe nasal septum fracture or coincident adjacent fractures, whereas closed reduction with a metal reduction instrument is invasive and complex relatively.³ We describe a new technique to treat type II depressed nasal bone fractures and correct the nasal functional and cosmetic appearance successfully.

METHODS

Patient Demographics

The study enrolled 20 consecutive patients who had a fresh nasal bone fracture (< 14 d) and was diagnosed with computed



FIGURE 1. (A) Schematic representation of the operation. One appropriate diameter and length Foley catheter were chosen, and the Foley catheter was advanced into nasal cavity and dilated. (Informed consent was obtained from each patient, and the study was performed in Declaration of Helsinki).

From the *Department of Plastic and Reconstructive Surgery, Shanghai 9th People's Hospital, Shanghai Jiao Tong University School of Medicine, Shanghai; †Department of Burn and Plastic Surgery, The Western Central Hospital of Hainan, Hainan; and ‡Department of Radiology, Huadong Hospital, Fudan University, Shanghai, China. Received June 16, 2023.

Accepted for publication June 25, 2023.

Address correspondence and reprint requests to Ke Xue, PhD, MD, Shanghai 9th People Hospital, Shanghai Jiao Tong University School of Medicine Shanghai, Shanghai China; E-mail: nanxuebeiyuan@126.com; Lin Qi, MD, PhD, Department of Radiology, Huadong Hospital Affiliated to Fudan University, Shanghai 200040, P.R. China; E-mail: Qilin86521@163.com

All the procedures in this study were approved by the Ethics Committee of Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine.

The authors report no conflicts of interest.

Copyright © 2023 by Mutaz B. Habal, MD

ISSN: 1049-2275

DOI: 10.1097/SCS.00000000000009568