Clin Oral Invest (2007) 11:303–306
DOI 10.1007/s00784-006-0091-y

SHORT COMMUNICATION

Intraoral osteotomies using piezosurgery for distraction in an infant with Pierre–Robin sequence

Max Heiland · Marco Blessmann · Philipp Pohlenz · Lei Li · Rainer Schmelzle · Felix Blake

Received: 14 May 2006 / Accepted: 28 November 2006 / Published online: 3 January 2007

Abstract Airway management in infants with Pierre Robin sequence (PRS) is often challenging. Advancement of the tongue base using mandibular distraction in neonates can avoid tracheotomy and is increasingly propagated. The osteotomies can be performed via intra- and extraoral approaches. Nowadays, for precise bone cutting, piezosurgical devices have been introduced in maxillofacial surgery, which we used for osteotomies via the intraoral approach in the 7-week-old patient presented in this study. Instead of a preoperative CT scan, a 3D data set was generated preoperatively in the operating theatre using a 3D C-arm system. After bilateral transcutaneous pin placement and osteotomies, a second 3D data set was acquired. Primary and secondary reconstructions clearly visualized the patient's bone despite the low level of ossification in newborns and the presence of a large amount of metal implants. In view of the low level of radiation exposure going along with the 3D C-arm imaging, a follow-up examination after removal of the distractors was performed. With this report of a 7-week-old female infant with the diagnosis of PRS, we want to extend the surgical and diagnostic armamentarium for the treatment of infants with PRS.

Keywords Pierre Robin sequence · Mandibular distraction · Piezosurgery · Cone-beam computed tomography · Airway management

M. Heiland (✉) · M. Blessmann · P. Pohlenz · L. Li · R. Schmelzle · F. Blake
Department of Oral and Maxillofacial Surgery, University Medical Center Hamburg–Eppendorf,
Martinistr. 52,
20246 Hamburg, Germany
e-mail: heiland@uke.uni-hamburg.de

Introduction

Established methods of airway management in infants with Pierre Robin sequence (PRS) has included prone position, nasopharyngeal tube, endotracheal intubation and tracheotomy [1]. Nowadays, advancement of the tongue base using mandibular distraction in neonates can avoid tracheotomy and is increasingly propagated [5, 12]. Although osteotomies via an intraoral approach have been reported even in neonates, the use of external incisions increases the younger the patients are [3, 15]. However, for precise bone cutting, piezosurgical devices have been introduced in maxillofacial surgery [14]. According to Stübinger et al. [13], this technique is thought to be superior to conventionally rotating instruments in close proximity to soft tissues like nerves. Therefore, we used piezosurgery for osteotomies via the intraoral approach in the 7-week-old patient presented in this study.

For preoperative radiological examination, cephalometric radiographs have been described with up to 50% of inadequate-quality cephalograms for accurate measurements in patients with PRS [4]. Other authors regularly perform preoperative CT scans in patients with PRS [11]. If a 3D data set is desired, sedation or general anaesthesia is often necessary, putting these patients with critical airways at an additional risk. Previously described for intraoperative 3D imaging after reduction in zygomaticomaxillary complex fractures, cone-beam computed tomography, which is nowadays a well established modality in dentomaxillofacial imaging [7], using a 3D C-arm system seems also promising for intraoperative imaging of infants [8]. Therefore, acquiring the 3D data set immediately before surgery in the OR, an additional anaesthesia for imaging in a radiological department or transfers during anaesthesia can be omitted.

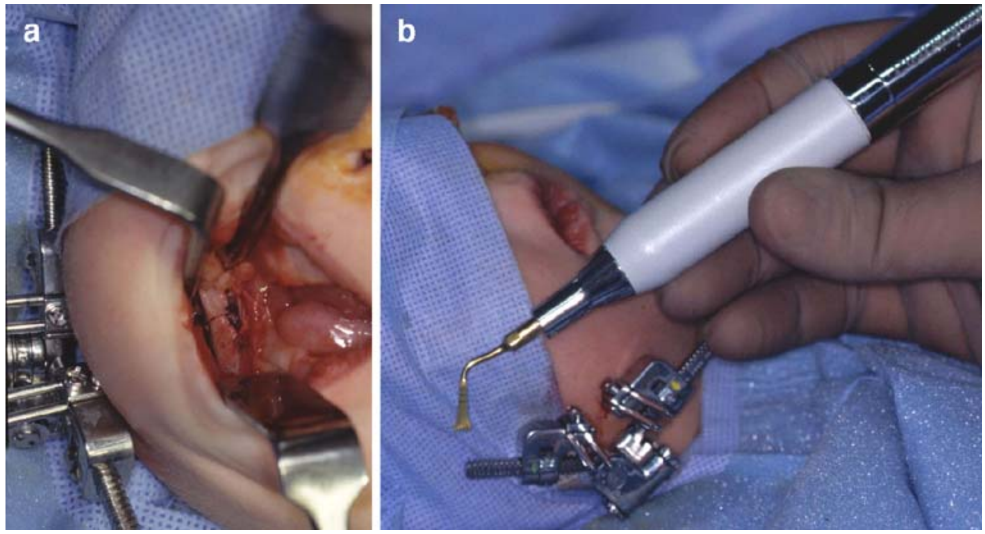

Fig. 1 **a** Intraoral osteotomy of the right mandibular ramus and transcutaneous placement of the distractor pins. As insert, a sharp-tipped saw was used with piezosurgical handpiece. **b** The angulations of the multi-directional distractor Multi-Guide II were adapted to the desired vector

With this case report, we want to extend the surgical and diagnostic armamentarium for the treatment of infants with PRS.

Case report

A 7-week-old female infant with the diagnosis of PRS was presented to the Department of Oral and Maxillofacial Surgery because of respiratory distress in the supine position. Since birth, recurrent decreased oxygen saturation to 75% with bradycardia occurred, demanding intensive care monitoring. However, sufficient respiration could be guaranteed by continuous positive airway pressure until the ninth day of life and later on by consequent positioning, resulting in the avoidance of tracheotomy. Because gavage was additionally necessary for at least 2 months, the patient was diagnosed as PRS group II according to Caouette-Laberge et al. [2]. To allow a normal development of the infant with an increasing range of action, a lengthening of the mandible by distraction going along with setting forward the tongue base was offered to the parents and performed after informed consent under general anaesthesia at the age of 55 days. Instead of a preoperative CT scan, a 3D data set was generated preoperatively in the operating theatre using the Arcadis Orbic 3D (Siemens Medical Solutions, Erlangen, Germany). The 3D C-arm was used in the “slow mode” performing 100 single projections in 60 s. Thereafter, the mandibular rami were exposed bilaterally via intraoral approach, and multi-directional distractors Multi-Guide II with 20-mm devices obtained from Leibinger micro implants (Stryker, Duisburg, Germany) were inserted transcutaneously using two 60-mm pins for bicortical fixation on each fragment. The osteotomies were performed transorally using a piezosurgery device manufactured by Mectron Medical Technology (Carasco, Italy, obtained from Rocker and Narjes, Cologne, Germany). As insert, a sharp-tipped saw was used (Fig. 1). For cutting the lingual cortical bone, a small chisel was used, preserving the lingual periosteum. The completeness of the osteotomies was checked by temporary activation of the distractors. A second 3D data set was acquired, and the operative result was immediately checked using secondary reconstructions generated with the help of the Syngo software (Siemens Medical Solutions; Fig. 2). The surgical procedure including two 3D X-ray examinations took 120 min. The distraction was started on the third postoperative day with 1 mm per day and continued for 10 days, followed by a consolidation period for 6 weeks. Because of postoperative swelling, the extubation was possible on the fifth postoperative day, and the intensive care was left on the tenth day. From the sixth postoperative day, the patient tolerated the supine position. Two months after the insertion of the distraction devices,

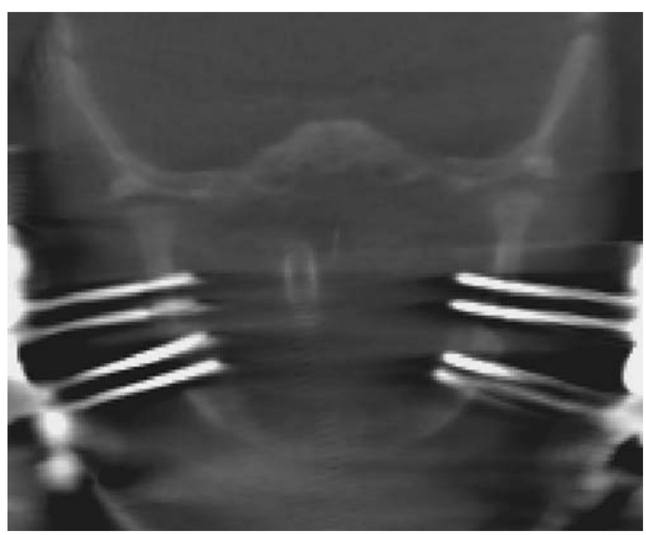

Fig. 2 Secondary coronal reconstruction clearly visualizes the patient’s bone, despite the low level of ossification in newborns and the presence of a large amount of metal implants

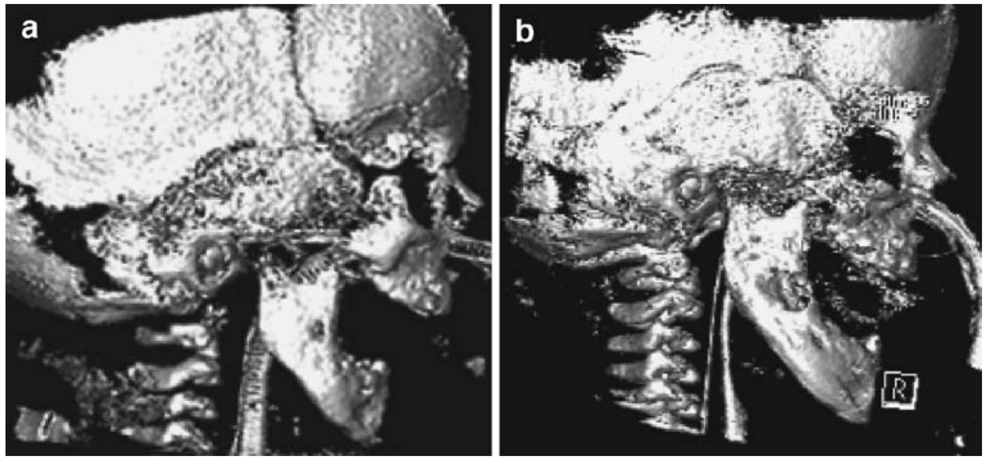

Fig. 3 **a** 3D SSDs of the first scan (before osteotomies) and **b** of the follow-up data set (after removal of the distractors)

they were removed, and a follow-up 3D data set was acquired intraoperatively.

Discussion

In infants with PRS, several surgical procedures have been described to manage the airway. Especially, tongue–lip adhesion has been advocated to relieve the airway obstruction and is propagated by Kirschner et al. [9] as first-line treatment of severe airway obstruction associated with PRS. In this paper, the authors emphasized external scars and long-term effects on the inferior alveolar nerve, on the primary and secondary dentition and on mandibular growth as disadvantages of mandibular distractions. However, with the here presented use of piezosurgery, that cuts mineralized tissue exactly and smoothly while soft tissue including nerves remain unharmed and which can be applied even in small surgical fields with limited view, the damage of the inferior alveolar nerve should be minimized [13, 14]. Furthermore, with intraoperative 3D imaging, a traumatic pin placement harming the inferior alveolar nerve or dental germs should be omitted or immediately corrected.

The intraoperative acquisition of 3D data sets proved to be uncomplicated and fast. Primary and secondary reconstructions clearly visualized the patient's bone despite the low level of ossification in newborns and the presence of a large amount of metal implants. Contrary to conventional radiographs with the intraoperative-acquired imaging data, the full spectrum of 3D reconstructions is available. The surface shaded displays (SSDs) reconstruction clearly demonstrates the lengthening of the mandible and the ossification of the newly formed bone, even after a time interval of only 2 months (Fig. 3). In view of the low level of radiation exposure going along with the 3D C-arm imaging, performing follow-up examinations during further procedures under general anaesthesia is justified and offers advantages compared with CT. According to our previously published study dealing with radiation exposure caused by the 3D C-arm, the patient with a total of three intraoperative scans and no conventional CT scan was exposed in total to half of the radiation going along with one conventional CT scan [10]. The applied imaging modality in this study seems also promising for data acquisition in infants with other craniofacial deformities in which 3D data sets proved to be of value [6].

References

1. Benjamin B, Walker P (1991) Management of airway obstruction in the Pierre Robin sequence. Int J Pediatr Otorhinolaryngol 22:29–37
2. Caouette-Laberge L, Bayet B, Laroque Y (1994) The Pierre Robin sequence: review of 125 cases and evolution of treatment modalities. Plast Reconstr Surg 93:934–942
3. Cohen SR, Simms C, Burstein FD (1998) Mandibular distraction osteogenesis in the treatment of upper airway obstruction in children with craniofacial deformities. Plast Reconstr Surg 101:312–318
4. Denny AD, Talisman R, Hanson PR, Recinos RF (2001) Mandibular distraction osteogenesis in very young patients to correct airway obstruction. Plast Reconstr Surg 108:302–311
5. Denny A, Kalantarian B (2002) Mandibular distraction in neonates: a strategy to avoid tracheotomy. Plast Reconstr Surg 109:896–904
6. Fisher DM, Lo LJ, Chen YR, Noordhoff MS (1999) Three-dimensional computer tomographic analysis of the primary nasal deformity in 3-month-old infants with complete unilateral cleft lip and palate. Plast Reconstr Surg 103:1826–1834
7. Guerrero ME, Jacobs R, Loubele M, Schutyser F, Suetens P, van Steenberghe D (2006) State-of-the-art on cone beam CT imaging for preoperative planning of implant placement. Clin Oral Investig 10:1–7
8. Heiland M, Schulze D, Blake F, Schmelzle R (2005) Intraoperative imaging of zygomaticomaxillary complex fractures using a 3D C-arm system. Int J Oral Maxillofac Surg 34:369–375
9. Kirschner RE, Low DW, Randall P, Bartlett SP, McDonald-McGinn DM, Schultz PJ, Zackai EH, LaRossa D (2003) Surgical airway management in Pierre Robin sequence: Is there a role for tongue–lip adhesion? Cleft Palate Craniofac J 40:13–18

10. Schulze D, Heiland M, Thurmann H, Adam G (2004) Radiation exposure during midfacial imaging using 4- and 16-slice computed tomography, cone-beam computed tomography systems, and conventional radiography. Dentomaxillofac Radiol 33:83–86
11. Sidman JD, Sampson D, Templeton B (2001) Distraction osteogenesis of the mandible for airway obstruction in children. Laryngoscope 111:1137–1146
12. Steinberg B, Fattahi T (2005) Distraction osteogenesis in management of pediatric airway: evidence to support its use. J Oral Maxillofac Surg 63:1206–1208
13. Stübinger S, Kuttenberger J, Filippi A, Sader R, Zeilhofer HF (2005) Intraoral piezosurgery: preliminary results of a new technique. J Oral Maxillofac Surg 63:1283–1287
14. Vercellotti T, De Paoli S, Nevins M (2001) The piezoelectric bony window osteotomy and sinus membrane elevation: introduction of a new technique for simplification of the sinus augmentation procedure. Int J Periodontics Restor Dent 21:561–567
15. Villani S, Brevi B, Sesenna E (2002) Distraction osteogenesis in newborns with Pierre Robin sequence. Mund Kiefer GesichtsChir 6:197–201