Case Report

Effective clinical outcome of a mandibular distraction device using three-dimensional CT with volume rendering in Pierre-Robin sequence

Filippo Cademartiri[1,2], Giacomo Luccichenti[1], Francesco Laganà[3], Bruno Brevi[3], Enrico Sesenna[3], Paolo Pavone[1]

[1] Department of Radiology, University of Parma, Parma, Italy
[2] Department of Radiology, Erasmus Medical Center, Rotterdam, The Netherlands
[3] Department of Maxxillo-Facial Surgery, University of Parma, Parma, Italy

Abstract. Computed Tomography (CT) with three-dimensional reconstructions was studied in cranio-facial deformities. The pre-operative and follow-up study of cranio-facial deformities can be performed with spiral CT. With this modality quantitative information can be provided in order to measure the entity of airway obstruction and the result of procedure affecting bone structures.

Key words: Three-dimensional; computed tomography; Pierre-Robin sequence; mandibular distraction device

Introduction

Congenital cranio-facial deformities cover a wide and complex range of diseases affecting especially newborn and infants. Complications are frequent and surgical procedure can be needed to avoid life threatening upper airway obstruction (1, 2).

Generally, tracheotomy is the solution of choice for refractory cases especially in emergency situations, but this procedure accounts for a great morbidity rate (3).

Mandibular distraction osteogenesis has recently acquired an important role in cranio-facial anomalies and, particularly, in the therapy of respiratory dysfunction related to cranio-facial deformities (4-6).

Computed Tomography (CT) with three-dimensional reconstructions have been already studied in cranio-facial deformities (7, 8). The pre-operative and follow-up study of cranio-facial deformities can be performed with spiral CT. In fact with this modality quantitative information can be provided in order to measure the entity of airway obstruction and the result of procedure affecting bone structures.

Herein is described a case of a newborn male affected by the Pierre-Robin Sequence (PRS) who was studied before and after the distraction osteogenesis procedure with Spiral CT and three-dimensional reconstructions.

Case description

B.L. (male) is an infant with PRS. At birth (March 2001) the clinical conditions were satisfactory: respiratory difficulties were controlled through medical procedures.

Fifteen days after birth he presented episodes of respiratory dysfunction. A naso-pharyngeal tube was placed to assure ventilation (Fig. 1a). Clinical condition worsened until it was decided to plan a surgical procedure.

At 2-month-age, he underwent bilateral mandibular distraction. The procedure was performed through the installation of two miniaturised iuxta-osseus lengthening devices applied at the mandibular distraction sites.

Distraction began 24 hours after surgical procedure. The rate of distraction was 1 mm per day for 15 days. Within 8 days, the patient was able to breathe without the naso-pharyngeal tube. After 15 days, he was fed per os reaching a normal diet (Fig. 1b).

Distraction was stopped at 15mm and the two distractors devices were removed 5 weeks later. The bone callus was of good quality with hyperplastic features (Fig. 1c).

The patient underwent unenhanced spiral CT with Somatom Plus 4 (Siemens – Forchheim – Germany) scanner before the installation and one week after the removal of the distraction osteogenesis device with the following parameters: collimation 2 mm, feed per rotation 2 mm (pitch 1), gantry rotation time 750 ms, and reconstruction index 1 mm.

Reconstructed axial images were sent to a workstation equipped with a software for 3D imaging and measurements (Vitrea 2.2, Vital Images, USA).

The airway volume before surgery along the obstructed tract was absent while after treatment there was an almost complete recovery of the airway volume

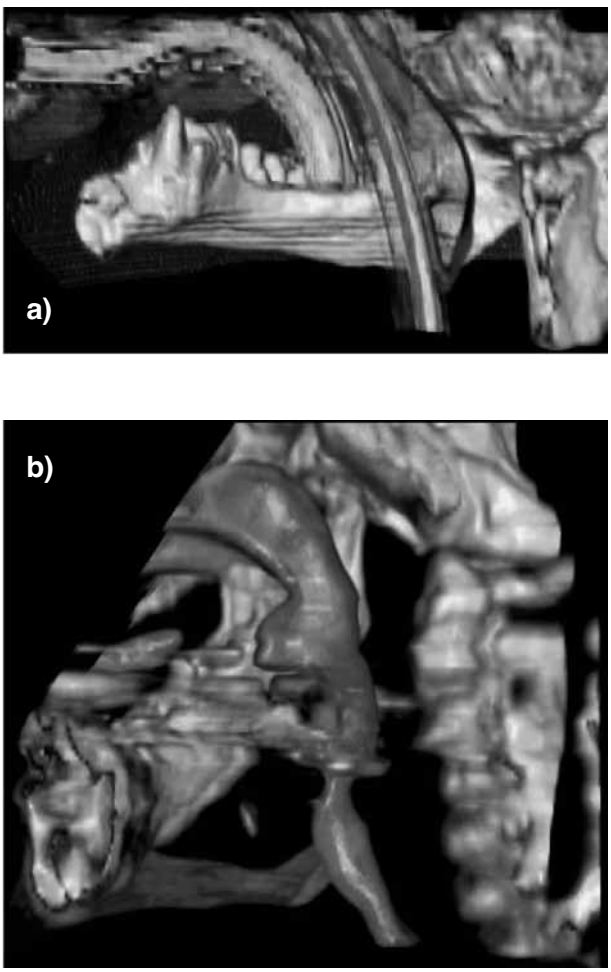

Figure 1. Three-dimensional Volume Rendering with coronal cut-plane before the positioning of the iuxta-osseus device (a). The patency of the airway is displayed. While the endo-tracheal tube descend from the naso-pharynx to the trachea, the patency of oro-pharynx and larynx stops just above the level of hyod bone (orange profile depicted around the endo-tracheal tube). In the background it is visible the right hypoplasic mandibular branch. Three-dimensional Volume Rendering with coronal cut-plane after the removal of the iuxta-osseus device (b). The patency of the airway is recovered (orange profile), and a few motion artefacts are present at the level of the oropharinx. The endo-tracheal tube has been removed and the distal airway is patent. Three-dimensional Volume Rendering with bone algorithm after the removal of the iuxta-osseus device (c). The irregular appearance of the left mandibular branch is due to the remodelling of the bone after the distraction procedure.

which became comparable to the one of the unaffected trachea distal to the obstruction (Fig. 1b).

After 12 months the child has grown normally, feed by himself and has no breathing problems.

Discussion

PRS is a congenital disorder that presents retromicrognathia, cleft palate and some degree of airway obstruction. The mouth cavity is smaller than usual, muscle development or co-ordination in the mouth may be poor and often the airway itself can be narrow.

The evaluation of craniofacial microsomia, of which PRS is one type, includes a thorough history and physical examination, photographic and cephalometric analysis, and three dimensional computed tomographic study. Family history of consanguinuity, intrauterine exposure to infection and toxins, and problems with delivery should be explored. Physical exam should focus on facial asymmetry as well as on isolated findings consistent with this syndrome. Photographs and cephalometry allow for monitoring of facial symmetry over time and aid in planning surgical approaches to individual patients.

There are two major and innovative features described in the management of this patient. The first concerns the application of a new internal iuxta-osseus innovative distraction osteogenesis device (Brevi-Sesenna distractor). The distractor was applied at 2-month-age to perform a bilateral mandibular distraction. The advantages of this type of distractor compared to the external ones can be resumed as follows: 1) a more effective application of the distraction force, with a consequent reduction of the procedure-related surgical trauma; 2) a more stable biomechanical structure, with a consequent reduction of intra and post-distraction complications (pseudo-arthrosis, pin loosening); 3) the low mass and volume of the device allows its application in newborn and infants; 4) and the reduction of mass and volume of the device allows to perform a minimally invasive procedure through a small surgical incision, with a consequent smaller scar. Until recently external device have been positioned for mandibular distraction osteogenesis (4-6).

The second feature concerns the use of spiral CT for the study of the patient and for the monitoring of therapeutic results. The scan protocol applied in this case was focused on the demonstration of airways and bone. Therefore contrast material was not used. The scan parameters should be decided based on: needed in-plane and through-plane spatial resolution (collimation and pitch), radiation exposure (mAs and pitch), needed temporal resolution (feed). In this case the pre-operative scan was kept as short as possible (short range to maintain thin collimation and low pitch), while in post-operative scan the range was enlarged.

One of the issues in this type of scan is motion. In this case the child was scan awake both pre and post-operatively. This is the reason why the pre-operative scan has a short range and the post-operative scan shows a few motion artefacts (mouth movements) at the level of the oro-pharynx. Nevertheless the overall result is not affected by those problems. In fact the recovery of airway patency is demonstrated.

Three-dimensional reconstructions were useful in order to display the configuration of the disease and the recovery of the correct mandibular position. Spiral CT data-set allowed to reliably assess the loss of airway's patency as well as its recovery after the distraction procedure.

Spiral CT with three-dimensional reconstructions provides accurate representation of patient' cranio-facial skeletons and virtually rules out the need for constructing physical models. This allows faster and more accurate treatment planning.

References

1. Byard RW, Kennedy JD. Diagnostic difficulties in cases of sudden death in infants with mandibular hypoplasia. *Am J Forensic Med Pathol* 1996; 17: 255-9.
2. Molina F, Ortiz Monasterio F. Mandibular elongation and remodeling by distraction: a farewell to major osteotomies. *Plast Reconstr Surg* 1995; 96: 825-40; discussion 841-2.
3. Myer CM, 3rd, Reed JM, Cotton RT, et al. Airway management in Pierre Robin sequence. Otolaryngol Head Neck Surg 1998; 118: 630-5.
4. Denny AD, Talisman R, Hanson PR, et al. Mandibular distraction osteogenesis in very young patients to correct airway obstruction. *Plast Reconstr Surg* 2001; 108: 302-11.
5. Klug CN, Millesi-Schobel GA, Millesi W, et al. Preprosthetic vertical distraction osteogenesis of the mandible using an

L-shaped osteotomy and titanium membranes for guided bone regeneration. *J Oral Maxillofac Surg* 2001; 59: 1302-8; discussion 1309-10.
6. Kawata T, Kohno S, Fujita T, et al. Transplantation of new autologous biomaterials into jaw cleft. J Int Med Res 2001; 29: 287-91.
7. Pozzi Mucelli RS, Stacul F, Smathers RL, et al. [3-dimensional craniofacial computerized tomography]. *Radiol Med* 1986; 72: 399-404.
8. Halimi P, Doyon D, Bekkali F, et al. Contribution of computerized tomography to the radiological study of craniofacial injuries. 34 cases. *J Neuroradiol* 1986; 13: 253-64.

Received: 3 October 2003
Accepted: 12 May 2004
Correspondence: Filippo Cademartiri
Viale Rustici, 2
43100 – Parma, Italy
Tel: 0039 0521 961833
E-mail: filippocademartiri@hotmail.com