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

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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.
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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.
which became comparable to the one of the unaffec-
ted 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 retro-
micrognathia, 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 cephalo-
metric analysis, and three dimensional computed to-
mographic study. Family history of consanguinity,
intrauterine exposure to infection and toxins, and pro-
blems with delivery should be explored. Physical exam
should focus on facial asymmetry as well as on iso-
lated findings consistent with this syndrome. Photo-
graphs and cephalometry allow for monitoring of fa-
cial symmetry over time and aid in planning surgical
approaches to individual patients.

There are two major and innovative features de-
scribed in the management of this patient. The first
concerns the application of a new internal iuxta-os-
seus innovative distraction osteogenesis device (Bre-
vi-Sesenna distractor). The distractor was applied at
2-month-age to perform a bilateral mandibular di-
striction. The advantages of this type of distractor
compared to the external ones can be resumed as fol-
lows: 1) a more effective application of the distraction
force, with a consequent reduction of the procedure-
related surgical trauma; 2) a more stable biomechani-
cal structure, with a consequent reduction of intra
and post-distraction complications (pseudo-arthro-
sis, 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 devi-
ce 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 (colla-
mation 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 po-
st-operatively. This is the reason why the pre-operati-
ve 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 re-
covery 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 di-
striction procedure.

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

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