Role of ultrasound in the active management of osteogenesis during compression–distraction osteosynthesis in children[☆]

A. Nadtochiy ^{a,b,*}

^a Department of Children's Maxillo-Facial Surgery, Central Scientific-Research Institute of Stomatology, Moscow, Russia

^b Department of Maxillo-Facial Surgery, St. Vladimir Children's Hospital, Moscow, Russia

KEYWORDS

Compression–distraction osteosynthesis;
Head and neck ultrasonography;
Mandibular hypoplasia;
Maxillo-facial surgery;
Musculo-skeletal ultrasonography.

Abstract *Purpose:* To determine whether distraction-induced regeneration (DR) can be optimized based on consecutive ultrasound findings during the course of compression–distraction osteosynthesis (CDO).

Materials and methods: Sixty-nine children ranging in age from 2 weeks to 15 years with congenital (37 patients) or acquired (32 patients) microgenia were treated with CDO. Radiological diagnostic studies included orthopantomography (OPG) and ultrasonography (US). The patients were divided into two groups: in group I a standard distraction rate (SDR) (1 mm/day) was used (33 patients, 47.8%); in group II (36 patients, 52.2%) individual distraction rates (IDR) were used based on US findings.

Results: DR was classified as normotrophic, hypertrophic, or hypotrophic based on US findings. In group I, 11 (33.3%) of the patients presented normotrophic DR, 8 (24.2%) had hypertrophic DR, and in 14 (42.4%), the DR was hypotrophic. In group II, all patients presented normotrophic DR at the end of the treatment.

Conclusion: US offers a unique opportunity to study DR development, and the results can be used to actively manage and optimize the osteogenesis process during mandibular CDO in children.

Sommario *Obiettivo:* Valutazione delle possibilità di ottimizzazione di sviluppo del tessuto di rigenerazione per distrazione (DR) basata sui segni dell'esame ecografico dinamico (USDE) nel corso dell'osteosintesi mediante compressione–trazione (CDO).

Materiali e metodi: Sessantanove bambini, di età compresa tra 2 settimane e 15 anni, affetti da microgenia (ipoplasia) congenita (37 pazienti) o acquisita (32 pazienti) sono stati trattati chirurgicamente con osteosintesi mediante compressione–distrazione (CDO). La diagnostica per immagini comprendeva l'ortopantomografia (OPG) e l'ecografia (US). I pazienti sono stati suddivisi in due gruppi: gruppo I – con rapporto di distrazione standard (SDR) pari a 1 mm/g – 33 pazienti (47,8%); gruppo II – con rapporto di distrazione individuale (IDR) – 36 pazienti (52,2%).

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* Department of Children's Maxillo-Facial Surgery, Central Scientific-Research Institute of Stomatology, ul. Timura Frunze, 16, Moscow 119992, Russia.

E-mail addresses: childface@mtu-net.ru, naggan@mail.ru

Risultati: Sono stati rilevati e descritti i segni ecografici equivalenti al tessuto di rigenerazione per distrazione (DR), normotrofico, ipertrofico e ipotrofico. Nel gruppo I il DR normotrofico si è ottenuto in 11 pazienti (33,3%), ipertrofico —8 (24,2%), ipotrofico —14 (42,4%). Nel gruppo II in tutti i pazienti si è sviluppato il DR normotrofico.

Conclusioni: L'esame ecografico dinamico USDE offre la possibilità di studiare lo sviluppo del DR; sulla base dei dati ecografici è possibile un trattamento attivo del processo di osteogenesi durante osteosintesi mediante compressione—distrazione (CDO) nei bambini.

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Introduction

Congenital or acquired hypoplasia of the mandible leads to esthetic problems and dental—maxillary disorders associated with various medical, psychological, and social problems [1–5]. The main methods used to correct mandibular hypoplasia are osteoplasty and compression—distraction osteosynthesis (CDO) [6–13]. The question of which is better naturally arises. Statistically, the chances for a positive outcome rarely exceed 60% with osteoplasty because this approach is associated with a high rate of immunological and inflammatory complications [3,5,10–13]. In contrast, CDO is associated with positive results in over 95% of all cases, and there is no risk of immunological problems because compression—distraction osteogenesis triggers native, organ-specific bone growth [14–20]. But some problems inevitably arise during CDO. Etiological factors, patient age, concomitant diseases and a host of other individual factors can significantly influence the rate of distraction-induced regeneration (DR), and these factors should all be taken into account during the distraction process. How can this be done *in vivo*? What diagnostic methods can be used to evaluate the early stages of DR development? And most importantly, can individual deviations in DR development be corrected in order to ensure regenerated bone with optimal characteristics? We found no answers to these questions in the currently available literature.

Materials and methods

Compression—distraction osteosynthesis was used to treat 69 children ranging in age from 2 weeks to 15 years with congenital (37 patients with Goldenhar and Franceschetti syndromes, Robin syndrome) or acquired (32 patients with traumatic or inflammatory temporo-mandibular joint arthrosis and ankylosis) microgenia, which was bilateral in 41 cases. These patients underwent a total of 69 distractions, and the formation and development of the DR was analyzed in all cases. Diagnostic imaging studies included orthopantomography (OPG) and ultrasonography (US) using special programs of application. OPG was performed once a week during the distraction period and once a month during the retention period until the compression—distraction device was removed. US was performed twice a week (every 3–4 days) during the distraction period, from the beginning of distraction, and once a month during the retention period until the CDD was removed. Therefore, each patient underwent dynamic US evaluation consisting of 7–10 exams.

Ultrasound examinations were performed in the B-mode with a Sonoline—Sienna scanner (Siemens) and a multi-frequency (5.0–7.5–9.0 MHz) linear transducer. Power and color Doppler studies were also used at the beginning of the study (in the first 17 patients),

but these modalities did not provide any useful information because the blood flow within the newly created capillaries of the DR is below the sensitivity threshold of Doppler systems. For this reason, Doppler studies were omitted in the latter part of the study. The scanning plane was perpendicular to the osteotomy line, i.e., the horizontal plane for the body of the mandible (0.5–1.0 cm above its base) and the frontal plane for the ramus (0.5–1.0 cm anterior to its posterior border).

The 69 patients were divided into two groups: group I was treated with a standard distraction rate of 1 mm/day (33 patients, 47.8%); in group II individualized distraction rates (IDR) were used (36 patients, 52.2%). In all patients from group I, passive radiological controls were carried out during the entire CDO cycle. At the end of CDO, OPG was used to analyze bone regeneration (BR) in terms of thickness and density, compared with the “maternal” bone around the osteotomy line. If the thickness and density of the regenerated bone was equal to that of the “maternal” bone, the BR was classified as “normotrophic”. If the regenerated bone displayed reduced thickness and reduced or equal density, compared with the “maternal” bone, the regeneration was considered “hypotrophic”. If the regenerated bone was equal in thickness with similar or increased density (compared with maternal bone) and there was pain or resistance to screwing movements during the distraction period, the BR was classified as “hypertrophic”.

The US images were then retrospectively analyzed to identify US findings associated with normotrophic, hypertrophic, and hypotrophic DR during the stages of primary formation and early development. (It is important to recall that at this stage, X-ray findings are negative and OPG reveals only the relative positions of the bone fragments.) Figs. 4–7 show examples of normotrophic, hypertrophic, and hypotrophic DRs based on differences in the echogenicity of the central zone of the DR (primary fibrous-tissue proliferation zone) and the peripheral zones (fibrous-tissue differentiation zones). It is important to note that these differences should be evaluated only during the distraction period.

Normotrophic BR was characterized by a hypoechoic central zone (sometimes with fine, moderately echogenic inclusions) and thin peripheral zones with moderate to high echogenicity (“light frame”). In hypotrophic BR, there was no distinction between the central and peripheral zones: the entire area appeared hypo- or anechoic. Hypertrophic DR was associated with a hypoechoic or intermediately echogenic central zone with multiple, large inclusions characterized by intermediate to high echogenicity) and wide peripheral zones with high echogenicity (“strong frame”). In all patients from group II, dynamic US data were used to adjust the distraction rate on an individual basis in order to achieve normotrophic BR.

Results

In group I, the DR was classified as normotrophic in 11 patients (33.3%), hypertrophic in 8 (24.2%), and hypotrophic in 14 (42.4%). In the vast majority of the patients with hypertrophic DR, the mandibular hypoplasia had been

Fig. 1 Congenital or acquired hypoplasia of mandible.

caused by arthrosis of the temporo-mandibular joint (7/11 patients—87.5%) whereas, congenital syndromes were responsible for the hypoplasia in 12/14 (87.5%) patients with hypotrophic DR.

In group II, 13 (36.1%) of the patients presented signs of hypertrophic DR in the early stages of the distraction process, and the distraction rate was thus increased. In nine others (25%), the DR initially appeared hypotrophic, and the distraction rate was therefore decreased. In all cases, the final result was normotrophic DR.

Discussion

Congenital or acquired hypoplasia of the mandible leads to esthetic problems and dental—maxillary disorders that are associated with a variety of medical, psychological, and social problems (Fig. 1) [1–5]. Modern maxillo-facial surgery makes it possible to normalize the mandible size using compression—distraction osteosynthesis (CDO) [3–5,9,13,21,22] (Fig. 2). Positive results can be expected in over 95% of all cases, and there are no immunological complications because CDO triggers native, organ-specific bone growth [14–20].

Fig. 2 Facial profile and esthetics: problems and results.

Fig. 3 Stages of compression—distraction osteosynthesis and the evolution of the regenerating tissue.

The onset and development of bone regeneration during CDO includes several stages (Fig. 3).

The first stage begins after osteotomy and fixation of the compression—distraction device and involves motionless compression of the bone fragments. This stage is characterized by the primary formation and proliferation of fibrous tissue. After 5–7 days of compression, the distraction stage begins. During this stage, fibrous-tissue formation and proliferation continues, and fibrous—osteoid transformation begins in the peripheral regions of the DR zone. As a result of these two different processes, the DR acquires a well-defined zonal structure characterized by fibrous-tissue formation and proliferation in the center and fibrous—osteoid transformation in the peripheral zones.

Conventionally, the mean distraction rate is around 1 mm/day. This rate is based on the mean velocity of capillary proliferation. The distraction stage lasts until the desired bone length is achieved and is followed by the retention stage of CDO. During this stage, fibrous—osteoid transformation of the proliferated fibrous tissue is completed and — in parallel — osteoid—osseous transformation begins in the peripheral (paraossal) parts of the DR. Upon

Fig. 4 Ultrasound images of the regenerating tissue.

Fig. 5 Clinical example of compression–distraction osteosynthesis.

completion of the ossification process, the DR loses its zonal structure. When this happens, the compression–distraction device should be surgically removed. The mean duration of the retention stage is 2 or 3 months.

It is interesting (and practically speaking, very important) that US allows visualization of the DR structure at each stage of CDO, as shown in the various panels of Fig. 4: the initial position of the bone fragments and the osteotomy line (A), evolution of the zonal structure of the DR (B, C) and DR mineralization (D). It is clear to the eye that the images in panels A and D are fairly similar. However, the clinical result is sufficiently impressive (Fig. 5), and it involves not only the patient's profile: the mandible lengthening normalized the positions of the root of the tongue and the hyoid bone (from their retroposition caused by mandibular hypoplasia), eliminating the risk of aspiration and allowing removal of the tracheostoma (Fig. 5).

X-ray examination – orthopantomography (Fig. 6) is routinely used for monitoring the results of CDO of the mandible. It can clearly reveal the relative positions of the bone fragments at the beginning of treatment, their subsequent displacement during the distraction stage, and DR

Fig. 6 X-ray monitoring of the compression–distraction process.

Fig. 7 Ultrasound monitoring of the compression–distraction process.

mineralization in the retention stage. However, this approach cannot provide an answer to the very important question: how is the DR developing during the distraction period? This phase is crucial because it is the initial phase that determines the condition of the future bone.

US examination of 33 patients (45 CDO procedures) subjected to CDO with a standard, constant distraction rate of 1 mm/day revealed normotrophic BR development in 11 patients (33.3%), hypertrophic development in 8 (24.2%), and hypotrophic development in 14 (42.4%) (Fig. 7). Most of the patients with hypertrophic BR had mandibular hypoplasia caused by traumatic or inflammatory arthrosis of the temporo-mandibular joint while the majority of those with hypotrophic BR had congenital defects associated with the Goldenhar and Franceschetti syndromes. The question is: how can these differences be explained when the distraction rate was identical in all patients (1 mm/day)?

We assumed that the differences observed in this group were a reflection of patient-specific rates of fibrous-tissue proliferation and differentiation, fibrous–osteoid

Fig. 8 Bone regeneration type is related to the equilibrium between individual osteogenesis activity and the distraction rate.

Fig. 9 Coordinating the distraction rate based on individual osteogenesis activity.

transformation and mineralization, hereafter referred to as *individual osteogenesis activity* (IOA). In other words, if two patients have different IOAs, e.g., a high IOA rate in patient A and a low IOA rate in patient B, and both are subjected to the same distraction rate, the former patient A will develop a hypertrophic DR/BR while the latter will develop a hypotrophic DR/BR. If this hypothesis is true, then the DR type should display correlation with the ratio of the IOA to the distraction rate. If the ratio is close to 1, the BR will be normotrophic. In cases characterized by relatively high IOA, a hypertrophic DR/BR develops, and in those with low IOA, the DR/BR will be hypotrophic (Fig. 8).

Therefore, normotrophic DR growth should be promoted by interventions that optimize the balance between the IOA and the distraction rate, and the simplest means of achieving this is to modify the individual distraction rate (IDR) (Fig. 9). If the two factors are equal in strength, the distraction rate might be kept on the same level. In cases with high IOA, the distraction rate might be increased, and in those with low IOA the distraction rate might be decreased. The final goal of these individual modifications of the distraction rate is the achievement of normotrophic DR in spite of the differences in IOA.

Fig. 10 Individual osteogenesis activity and distraction rate correction.

Fig. 11 Clinical confirmations.

The crucial point of this technique lies in IOA monitoring. This can be effectively achieved with an US examination performed after 3–4 days of CDO at a standard distraction rate of 1 mm/day. US signs of normotrophic DR indicate that the IOA is normal (and the constant distraction rate should be maintained); US signs of hypotrophic DR indicate a low IOA (and the distraction rate should be decreased); US signs of hypertrophic DR indicate a high IOA (and the distraction rate should be increased, Fig. 10).

These hypotheses were extensively tested and confirmed by our study of 49 BR zones in 36 patients. In this study, US monitoring and distraction-rate correction were used on a permanent basis in a way that hypertrophic sonographical findings triggered positive corrections of the distraction rate, while hypotrophic findings triggered the negative corrections. The results of these efforts were excellent, with normotrophic DR consistently achieved in all patients (Fig. 11).

Conclusion

US offers a unique opportunity to evaluate DR development and define the patient's IOA status. Based on the findings of ultrasound monitoring, active management of the distraction rate can promote normotrophic bone regeneration during the course of CDO.

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