Original Article - Retrospective Study

Perpendicular serial maxillary distraction osteogenesis in cleft lip and palate patients

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Leena P. Ylikontiola[1], George K. Sándor[1,2], Virpi Harila[3]

[1]Department of Oral and Maxillofacial Surgery, Medical Research Centre, Oulu University Hospital, Institute of Dentistry, University of Oulu, [3]Department of Oral Development and Orthodontics, Oral Health Sciences Research Group, Medical Research Centre, Oulu University Hospital, University of Oulu, Oulu, [2]BioMediTech, Institute of Bioscience and Technology, University of Tampere, Tampere, Finland

Address for correspondence:
Prof. George K. Sándor, University of Oulu, Aapistie 5, Office No: 2126B, 90014 Oulu, Finland.
E-mail: george.sandor@oulu.fi

ABSTRACT

Background: Cleft lip and palate patients often have a retruded maxilla with a severely narrowed deficient maxillary arch. This report aims to describe the management of severe maxillary retrusion and constriction in cleft lip and palate patients using distraction osteogenesis applied in serial sequence in two directions perpendicular to each other. **Materials and Methods:** Two adult male cleft lip and palate patients were treated with maxillary distraction osteogenesis in two stages. In the first stage, surgically assisted rapid palatal expansion with a tooth-borne device was performed to significantly expand the maxillary arch in the transverse dimension. After the teeth were orthodontically aligned, the horizontal distraction of the maxilla was made by two internal maxillary distraction devices. **Results:** In the first patient, the maxilla was initially widened by 11 mm and then distracted forward by 20 mm. Despite the breakage of the shaft of one of the two distractors at the end of distraction, a satisfactory occlusion was found at the time of distractor device removal. The maxillary position has remained stable through 8 years of follow-up. In the second patient, the palate was widened by 14 mm and the maxilla was distracted forward by 22 mm. The maxillary position has remained stable through 3 years of follow-up. **Conclusion:** Sequential serial distraction of maxilla in two planes perpendicular to each other is a safe and stable approach for the treatment of cleft lip and palate patients with severe transverse and anteroposterior discrepancies.

Keywords: Cleft lip and palate, maxillary distraction osteogenesis, palate expansion

INTRODUCTION

Patients with cleft lip and palate have the tendency to develop maxillary hypoplasia with the need for maxillary anteroposterior advancement. They also often have severe loss of palatal transverse dimension due to scarring and altered palatal morphology.[1,2] Since standard osteotomies are especially prone to relapse in cleft patients, clinicians have turned to distraction osteogenesis to minimize the relapse.[3]

Distraction osteogenesis as it applies to the midface is not a new concept.[4] Dentists have used techniques that involve the application of both tensile and compressive forces to the bones of the craniomaxillofacial skeleton for almost 300 years. According to Balaji, Fauchard described the use of an expansion arch as early as 1728, using a custom-made metallic arch applied to the crowded maxillary dentition, to widen the arches to a more physiologic form.[5] Wescott attempted to correct a crossbite by placing two double clasps on the maxillary bicuspid teeth and a telescopic bar to apply transverse force.[5] Similarly, Angell expanded a maxillary arch by a transverse jack-screw and clasps upon the bicuspid teeth.[6] Goddard is

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credited with standardization of the palatal expansion protocol with activation twice daily for 3 weeks followed by a period of stabilization.[7] Modern clinical distraction osteogenesis was introduced in the long bones by Illizarov.[8] Distraction of the facial bones developed quickly once McCarthy applied the concept to mandibular lengthening in 1992.[9] This led to an explosion of clinical and research activity in the field of craniomaxillofacial distraction osteogenesis over the past two decades.[10]

Cleft lip and palate patients may require significant distraction of their maxillae with the advancement of their midface at one or more Le Fort levels and possibly in more than one plane with palatal expansion. Maxillary advancement using traditional osteotomies may place these patients at risk not only for skeletal relapse[11] but also for the development of velopharyngeal insufficiency.[12] It has been reported that this debilitating complication may be avoided for some of these patients if distraction osteogenesis techniques were used to advance the maxilla.[3,11,13]

When there is a severely narrowed maxilla, then surgical widening of the maxilla can be used in order to correct for transverse maxillary deficiency in patients with a fused midpalatal suture.[14-17] This procedure has been termed surgically assisted rapid palatal expansion (SARPE). While it predates all other distraction osteogenesis procedures performed in the midface, SARPE is often forgotten in the classification of midfacial distraction.[4] The SARPE procedure is very useful in helping to produce a stable widening of the maxillary arch, even when the arch is significantly constricted, as long as the necessary surgical-orthodontic treatment principles are used.[18-20] As in other forms of distraction osteogenesis, the distraction of the midpalatal suture permits a larger correction than orthodontic treatment alone could achieve.[21-23] This form of distraction osteogenesis can be safely employed early in surgical-orthodontic treatment, sometimes as the first of a sequence of surgical procedures.[24]

The risks associated with distraction osteogenesis of the midfacial structures are similar to the risks encountered with traditional osteotomies. Careful preoperative planning of the vectors of distraction is essential to ensure that there is no convergence and that the distracted segment will advance fully in the desired direction without interference from surrounding bony structures or teeth.[4]

The purpose of this report is to describe the management of severe maxillary retrusion and constriction in cleft lip and palate patients by using distraction osteogenesis applied in a serial sequence in two different directions and to provide some long-term follow-up.

MATERIALS AND METHODS

Two adult male patients with cleft lip and palate were referred for management of their severe malocclusions. In the case 1, the patient was a 20-year-old male who had received a sequence of cleft lip and palate care including an alveolar bone graft at the age of 13 years. The patient was noted to have midfacial hypoplasia with maxillary retrusion [Figures 1-3]. In addition, there was a severe transverse deficiency, complete cross-bite, and severe crowding due to tooth-size jaw-size discrepancy [Figures 4 and 5].

Distraction of the maxilla was planned in two stages. At first, a SARPE with a tooth-borne device [Figure 6] was used to distract the maxillary arch in the transverse dimension in order to expand the palate over a 2 weeks period [Figures 7 and 8]. Once orthodontic tooth alignment was completed [Figures 9-11], the distraction hardware was fitted onto a stereolithic skull and their vector alignments were checked [Figures 12 and 13]. Horizontal distraction of the maxilla was performed with a Le Fort I osteotomy using two Synthes internal maxillary distraction devices (Synthes, Oberdorf, Switzerland) over a period of 2 weeks [Figures 14-22].

The second case was a 32-year-old male who presented with severe midfacial retrusion and transverse deficiency [Figures 23-26]. The patient's management also included transverse maxillary distraction by SARPE to expand the severely constricted maxillary arch followed by orthodontic alignment of severely malpositioned teeth [Figure 27]. Like the first case, the time to align the teeth between the two phases of perpendicular distraction was 1 year. The patient was also treated with a Le Fort I level osteotomy and placement of two Synthes internal maxillary distractors [Figures 28-36].

Both patients had their distractors removed 3 months following the completion of anteriorly directed distraction and the distraction hardware was replaced by four L-shaped plates to provide internal fixation. Orthodontic treatment was completed, necessary dental implants were placed, then restored and the patients were followed-up at regular intervals. The records of the patients including chart entries, photographs, radiographs and dental casts were analyzed and the details were reported. The case series was approved as a retrospective study by the Ethical Committee of the Oulu University Hospital and the data information were treated with the principles of the Helsinki Declaration.

RESULTS

In the case 1, the maxilla was initially widened by 11 mm and distracted forward by 20 mm. Despite the breakage of the shaft of one of the two distraction devices noted at the end of distraction [Figure 18], a satisfactory occlusion was noted at the time when the distraction devices were removed and replaced with miniplates.

The maxillary position in case 1 has remained stable over 8 years of follow-up with a 1 mm upward migration at "A" point. In case 2, the palate was widened by 14 mm and the maxilla was distracted forward by 22 mm. Dental implants were placed into the newly formed bone in the wake of the transversely distracted maxilla with no need for further bone grafting or augmentation of the ridge [Figure 30]. The maxillary position has relapsed 2 mm posteriorly at "A" point over 3 years of follow-up. Despite the long maxillary advancements, neither patient in this series developed any speech complications such as velopharyngeal insufficiency.

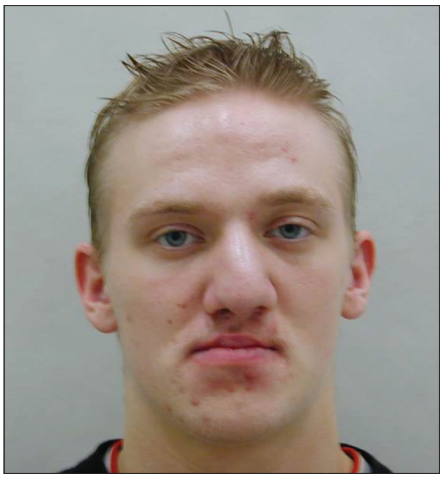

Figure 1: Preoperative frontal photograph of case 1

Figure 2: Preoperative lateral photograph of patient

Figure 3: Preoperative photograph of severely malaligned and malpositioned teeth with severe transverse maxillary deficiency

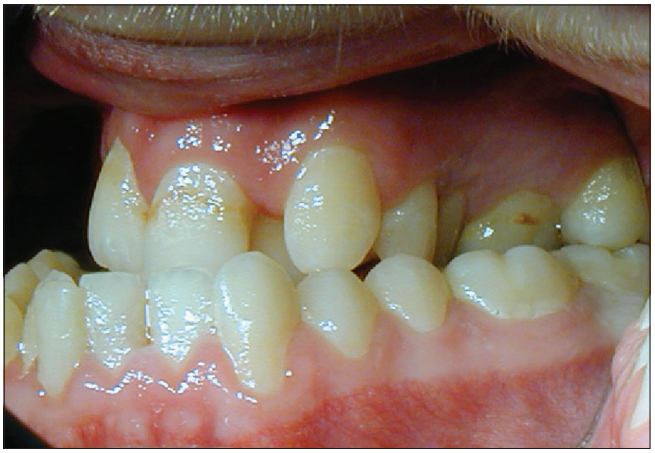

Figure 4: Preoperative left lateral buccal segment view of occlusion

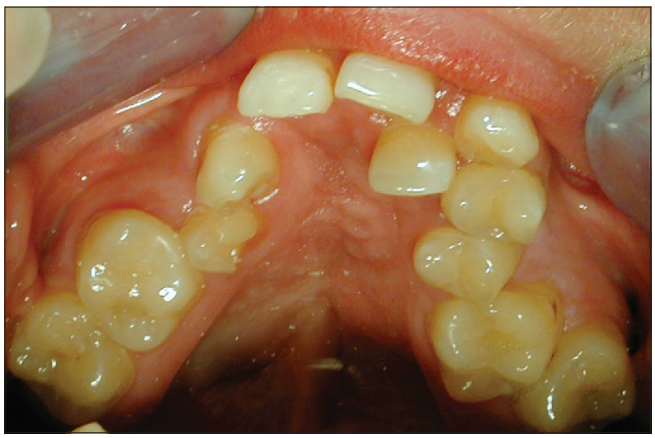

Figure 5: Preoperative occlusal view showing severely constricted maxillary arch

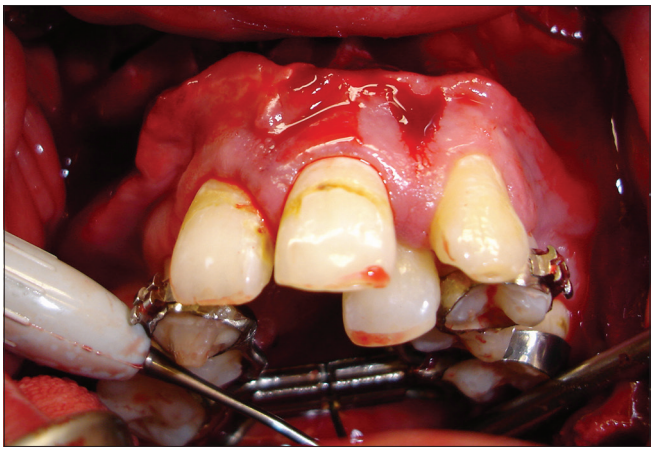

Figure 6: Maxillary tooth-borne device being activated to initiate transverse maxillary distraction

DISCUSSION

The midfacial deformities seen in cleft lip and palate patients include transverse maxillary deficiency, midfacial retrusion, and significant alveolar cleft defects which have been treated in the patients reported in this series by serial distraction osteogenesis in two different planes of space. Distraction osteogenesis offers several advantages over conventional osteotomies in the treatment of cleft lip and palate patients including postoperative stability. There is a reduced tendency for significant relapse following distraction of the maxilla than after traditional maxillary osteotomies.[25-27] The

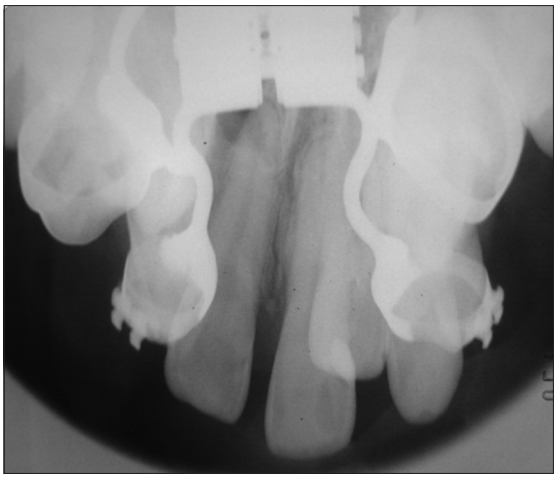

Figure 7: Occlusal radiograph taken at the start of maxillary transverse distraction

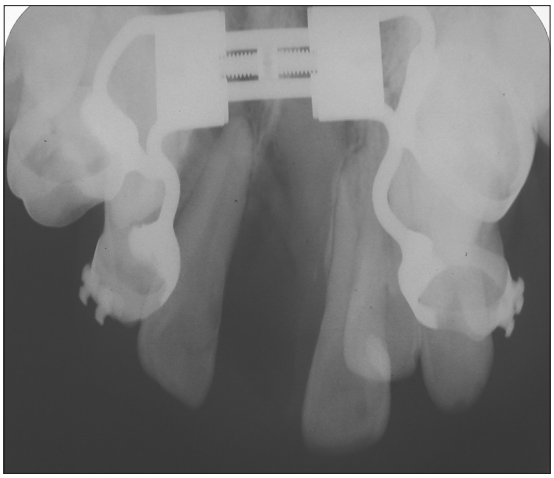

Figure 8: Occlusal radiograph taken at the end of maxillary transverse distraction. Note the large diastemma that has been produced in the midline

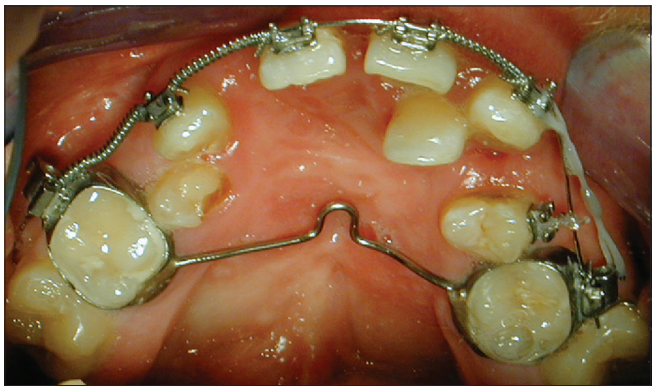

Figure 9: Maxillary occlusal view photograph showing teeth being aligned in the maxilla following its transverse distraction

Figure 10: Photograph showing retrusive maxilla prepared for advancement

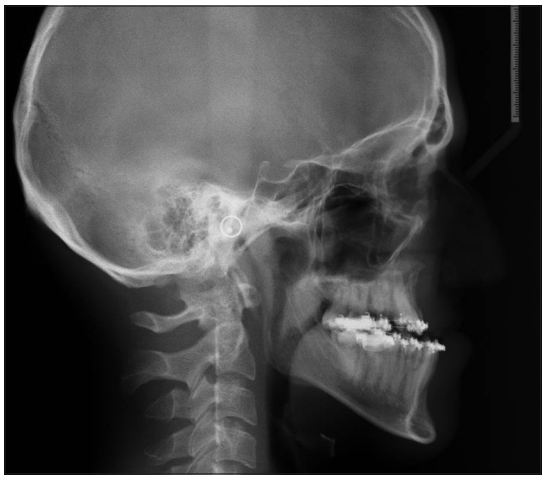

Figure 11: Lateral cephalometric radiograph just prior to maxillary anterior distraction

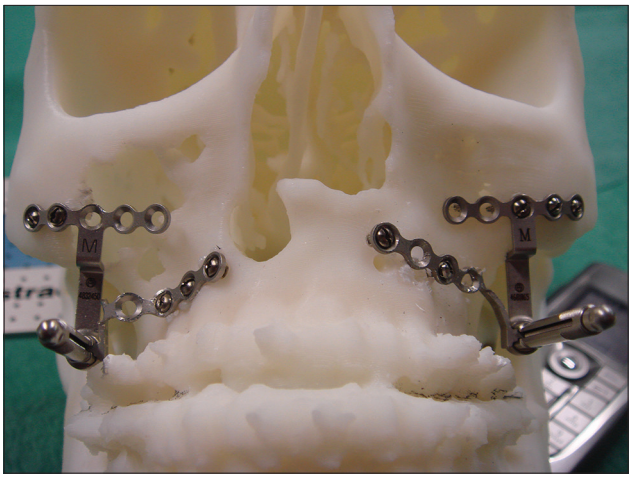

Figure 12: Maxillary distractor footplates are prebent and fitted and secured with screws to stereolithic skull of patient

soft tissue changes associated with maxillary advancement may be superior following distraction osteogenesis when compared to traditional Le Fort I level advancement surgery.[28-30] Maxillary distraction may help correct obstructive sleep apnea syndrome.[31] Avoidance of deterioration in velopharyngeal function in patients at risk for the development of velopharyngeal insufficiency is another advantage of maxillary distraction.[3,11,13,32]

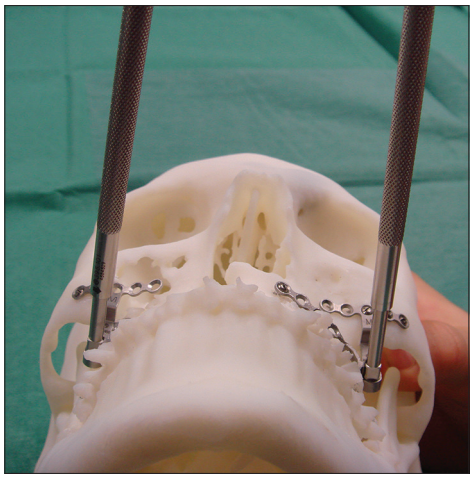

Figure 13: Alignment rods are attached to the distractors installed on stereolithic skull to check alignment

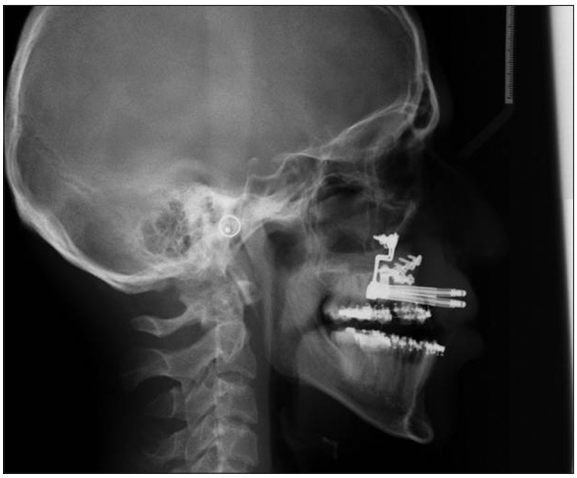

Figure 15: Postoperative lateral cephalometric radiograph showing maxillary distractors at the beginning of distraction

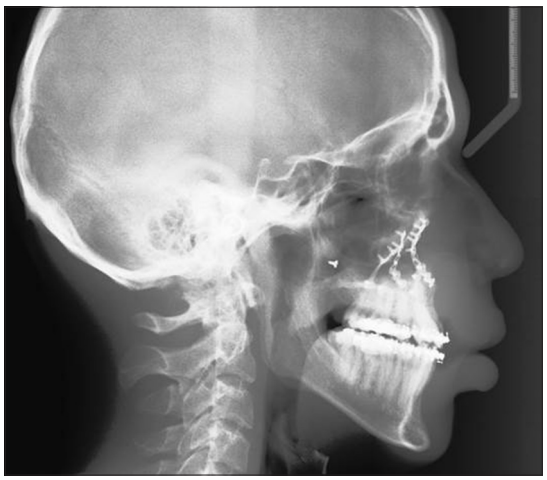

Figure 17: Lateral cephalometric radiograph taken after distractor removal and replacement by 4 L-shaped internal fixation plates

Internal distraction hardware[33,34] has been developed for Le Fort I level osteotomies [Figure 12] in embodiments designed

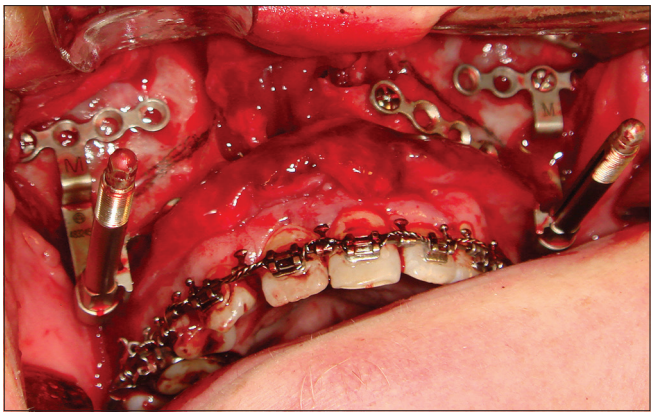

Figure 14: Maxillary distractors fitted at time of Le Fort I osteotomy

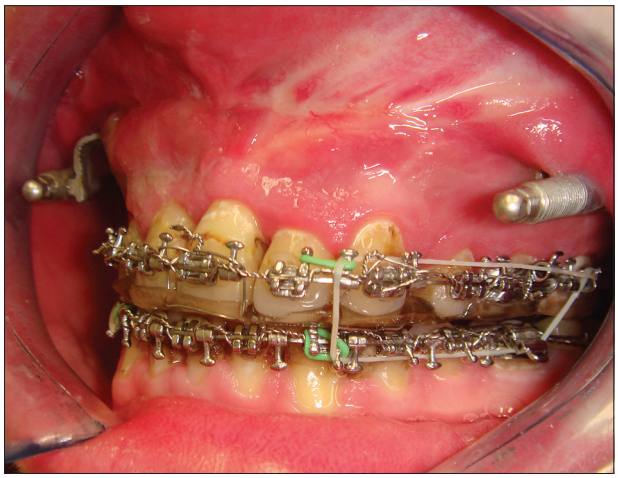

Figure 16: Maxilla distracted into its new occlusal relationship. The distractors are kept in place for 3 months following the attainment of the final maxillary position as retention devices

Figure 18: Broken distractor rod with a crack in its shaft noted at the time of distractor removal. This breakage seemed to have no influence on the distraction outcome

to be used submucosally and subcutaneously. The selection of a specific device is determined by the specific goals of the distraction procedure, anatomical constraints and the amount

Figure 19: Postoperative frontal photograph of patient

Figure 21: Postoperative right lateral occlusal view

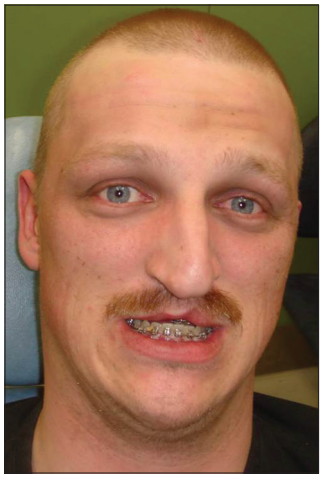

Figure 23: Preoperative frontal photograph of case 2

Figure 20: Postoperative anterior view of occlusion with class I molar relationship and no cross-bite

Figure 22: Postoperative left lateral occlusal view

Figure 24: Preoperative anterior view of occlusion of constricted and retrusive maxilla after preliminary orthodontic alignment

of room available to accommodate placement of the hardware. A stereolithic skull reconstructed from a three-dimensional computed tomography scan can aid in the planning of such osteotomies by permitting preoperative selection and bending of plates, thus reducing expenditures on distraction hardware and operating room time [Figure 12]. Preoperative planning also ensures that a certain configuration and arrangement of the selected distraction hardware will actually produce the desired vectors of distraction [Figures 13 and 14]. Future surgical workflow will allow the use of computer designed distractors with footplates that fit exactly to the surfaces of their desired location on the craniomaxillofacial skeleton.[35]

Figure 25: Right lateral view of buccal segment showing severely retrusive maxilla and complete cross-bite

Figure 26: Left lateral view of buccal segment showing severely retrusive maxilla and complete cross-bite

Figure 27: Anterior view of occlusion after transverse distraction of maxilla increasing the palatal width by 14 mm

Figure 28: Predistraction lateral cephalometric radiograph showing severe maxillary retrusion

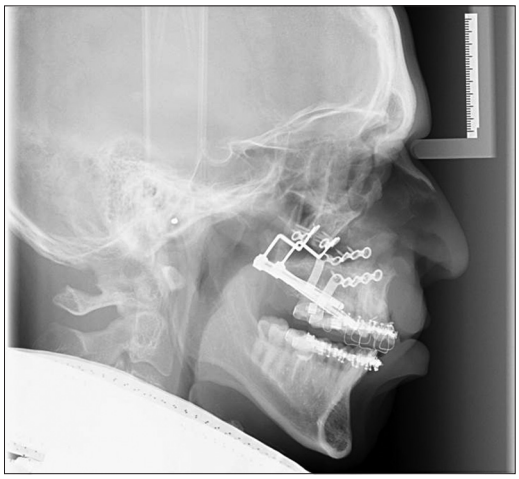

Figure 29: Lateral cephalometric radiograph showing maxilla with distraction devices and maxilla partway advanced

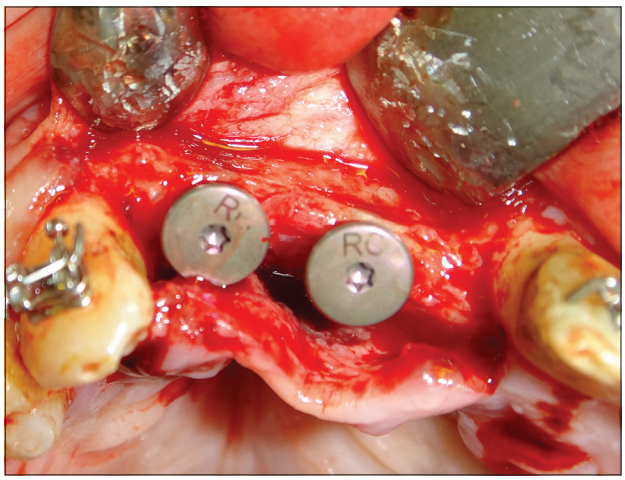

Figure 30: Dental implant being placed into generously wide anterior maxillary ridge left by the wake of bone produced by maxillary transverse distraction procedure without the need for bone grafting or augmentation of the ridge

While external devices are used in certain cases, those in favor of internal devices point out that such devices can be worn out of plain sight and have minimal impact on the daily activities of the patient.[36,37] From a patient perspective, internal devices are preferred although reports indicate that there is a transient drop in self-esteem even with internal distractor wear. The self-esteem improves following distractor removal.[38] Internal devices are often unidirectional so that distraction may be possible in only

Figure 31: Postretention lateral cephalometric radiograph with distractors having been removed 3 months following the completion of distraction. Internal fixation devices have been applied for long-term retention, and dental implants are in place with anterior incisal alignment normalized

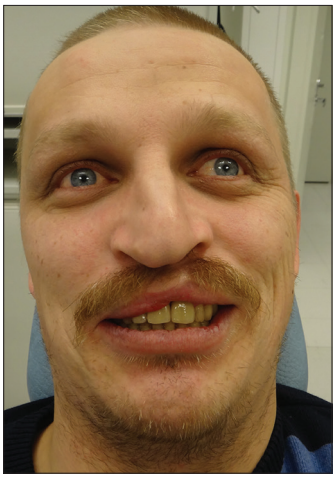

Figure 33: Postoperative frontal facial photograph of case 2 showing improved facial esthetics with new position of anteriorly distracted maxilla with improved subnasal support

Figure 35: Postoperative right lateral buccal segment view of occlusion with no cross-bite

Figure 32: Posttreatment panoramic radiograph showing restored dental implants in position

Figure 34: Postoperative anterior view of the occlusion and dental implant restorations

Figure 36: Postoperative left lateral buccal segment view of occlusion with no cross-bite

one plane of space. Often two internal devices must be used simultaneously on either side of the maxilla in order to obtain a symmetrical distraction. The cost associated with the use of two appliances must be borne in mind. Metal internal distraction devices are most rigid but require removal following the distraction process. Removal of metallic distraction devices, as with other hardware, can be difficult and complicated. Resorbable distraction devices that do not require removal have recently

become available.[39,40] Just as the skin around the fixation pins of an external distraction device can become infected, the tissues surrounding the transcutaneous or transmucosal distraction rods of the internal devices can also become infected. None of these complications occurred in the two patients reported in this series.

The sequential serial distraction of the maxilla in two planes perpendicular to each other is a safe, stable and controlled approach for the treatment of cleft lip and palate patients with severe transverse and anteroposterior discrepancies [Figures 5 and 9]. In the current protocol, the maxilla should be first distracted in the transverse plane, followed by orthodontic alignment of the malpositioned teeth. The time interval between the two phases or directions of distraction is the time taken to level and align the teeth, in both cases in this series this was 1 year. The maxilla is then distracted in the anteroposterior plane. Having the teeth aligned allows determination of the anteroposterior distraction end points more reliably.

The authors recognize that this is a case series of two and that the conclusions are therefore limited. The authors plan to continue this protocol and report further cases once appropriate. In the future, serial perpendicular distraction may be compared to simultaneous perpendicular distraction protocols.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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