CASE REPORT

# Mandibular distraction osteogenesis for treatment of extreme mandibular hypoplasia

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Distraction osteogenesis is an important option for the treatment of mandibular hypoplasia. This case report describes the treatment of a girl with mandibular hypoplasia and sleep apnea. She had a tracheostomy, followed by orthodontic treatment, including extraoral mandibular distraction osteogenesis to open the airway. Follow-up records 4 year 8 months after the distraction osteogenesis are presented. (Am J Orthod Dentofacial Orthop 2007;132:848-55)

The use of distraction osteogenesis (DO) to correct extreme mandibular hypoplasia has become fairly common. This intervention is intended not only to improve oral and craniofacial morphology but, in severe cases, to also maintain an open airway and improve the patient's general condition.

An absolute indication for DO in these patients is obstructive sleep apnea syndrome (OSAS). During sleep, the patient has difficulty maintaining an open airway, and hypopnea or apnea occurs. If the value of arterial oxygen saturation measured by pulse oximetry is very low, OSAS can result in general growth retardation;[1] in the past, such a patient might be treated with a tracheotomy. Correcting the mandibular hypoplasia and expanding the airway can prevent growth retardation and avoid the need for a prolonged tracheostomy with its cumulative morbidity and reduction in quality of life.[2]

This case report describes the treatment of a patient with mandibular hypoplasia due to perinatal infection of the temporomandibular joint and with OSAS as a consequence. Treatment included a tracheostomy, followed by mandibular distraction osteogenesis.

## DIAGNOSIS AND ETIOLOGY

The patient, a young girl, came to the orthodontic clinic at the Academic Center for Dentistry in Amsterdam in 1996. She had severe OSAS, an Angle Class II Division 1 malocclusion, and severe mandibular retrognathia. Overjet was 6 mm, and overbite was –1.5 mm. The midline of the maxilla was shifted 4.5 mm to the left of the midline of the mandible. Maxillary left first deciduous incisor, maxillary left second deciduous incisor, and mandibular left first deciduous molar were decayed. Dental age was 7 years 6 months (Fig 1).

The patient was born after a normal pregnancy. However, 17 days after birth, staphylococcus osteomyelitis of both mandibular condyles and hip joints was diagnosed and treated.

When she first came to the orthodontic department, she was just over 7 years old and had mandibular hypoplasia and retarded general growth. OSAS was diagnosed on the basis of polysomnography. She suffered from a severe form of hypercapnia and hypoxemia. Her arterial oxygen saturation measured by pulse oximetry was about 90%, with a minimum of 85%, probably because of compromised pharyngeal airway space; this could explain the growth retardation.

One month after referral to the orthodontic department, at age 7 years 5 months, she had a tracheostomy to keep the airway free. After that, her respiratory condition improved, and her general growth, measured by stature, accelerated (Fig 2). The tracheostomy was performed in 1996, and treatment of OSAS has changed since then.

## TREATMENT OBJECTIVES

After a period of catch-up growth, DO was planned to correct the mandibular retrognathism and improve the airway space so that the tracheostomy could be undone. The DO was also intended to solve the Class II relationship, normalize overjet and overbite, and improve facial appearance. Before the DO, overjet was 7 mm, and overbite was 2 mm. The midline of the maxilla was shifted 5 mm to the left compared

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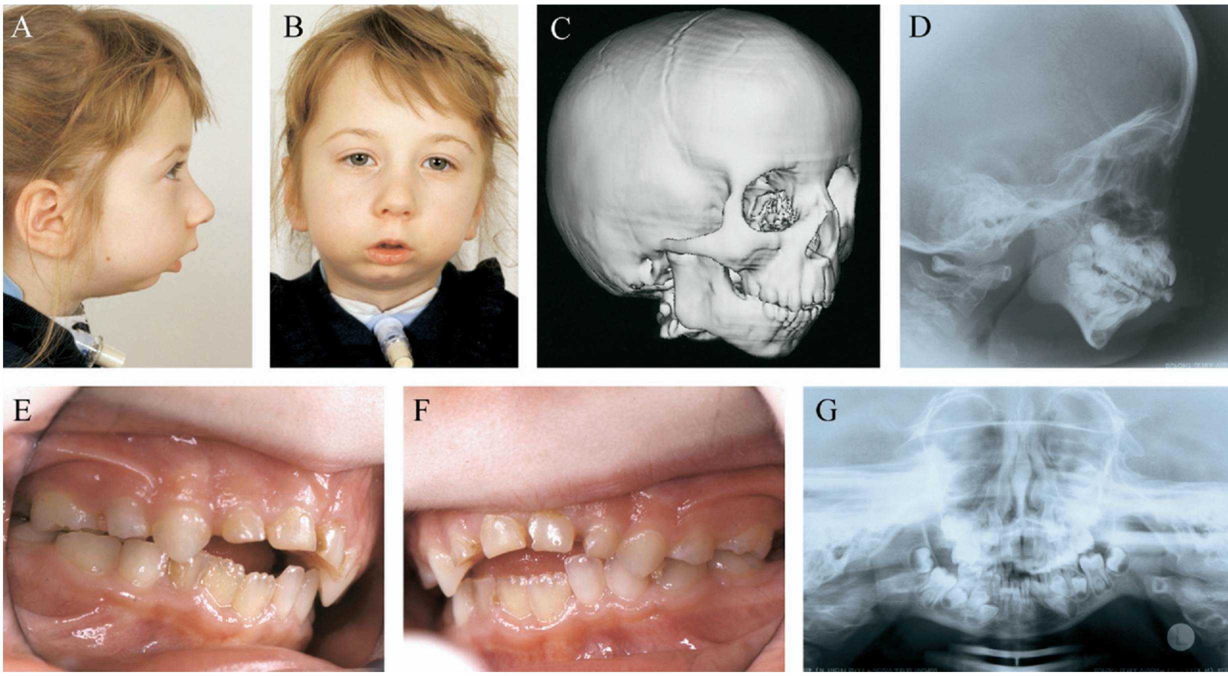


**Fig 1.** Posttracheostomy records at 7 years 6 months: **A** and **B,** facial photographs; **C,** 3-dimensional computed tomography scan; **D,** cephalometric radiograph; **E** and **F,** intraoral photographs; **G,** panoramic radiograph.

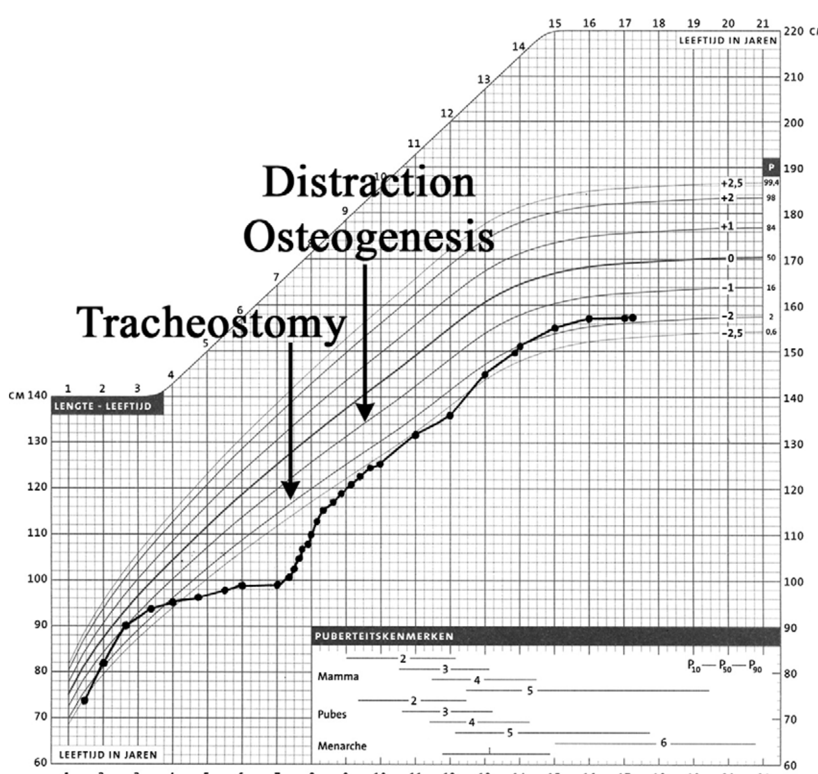


**Fig 2.** General growth chart.

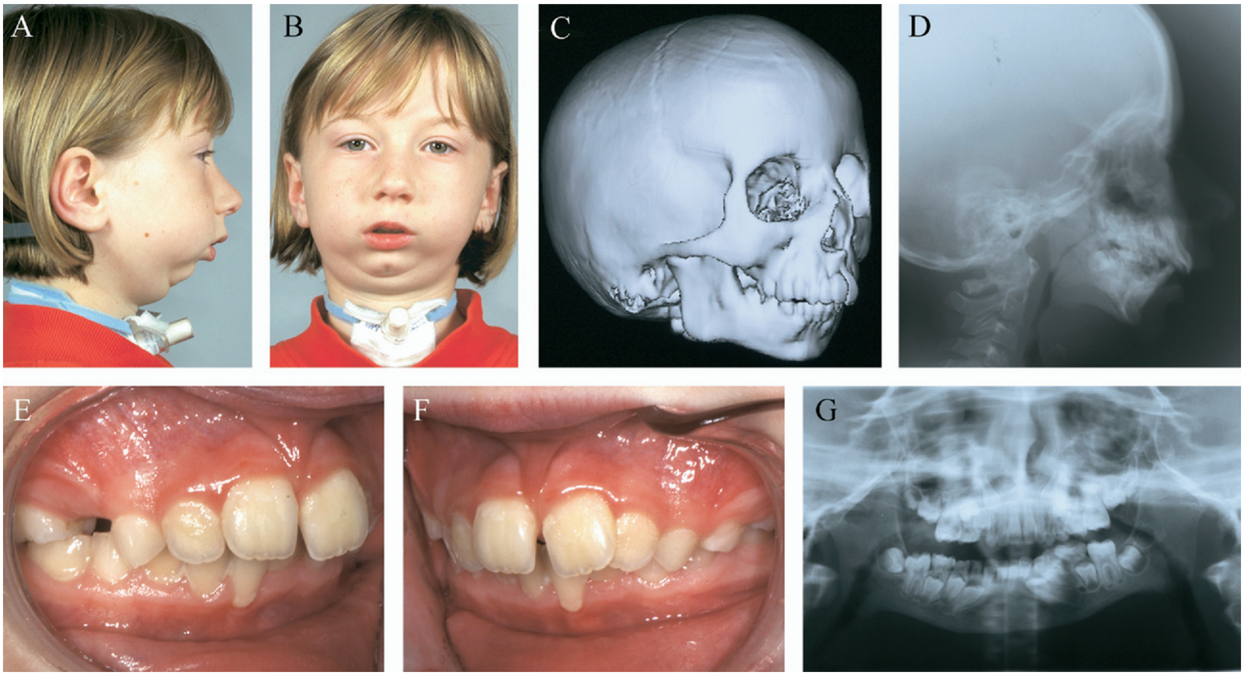


**Fig 3.** Predistraction records age 9 years: **A** and **B,** facial photographs; **C,** 3-dimensional computed tomography scan; **D,** cephalometric radiograph; **E** and **F,** intraoral photographs; **G,** panoramic radiograph.

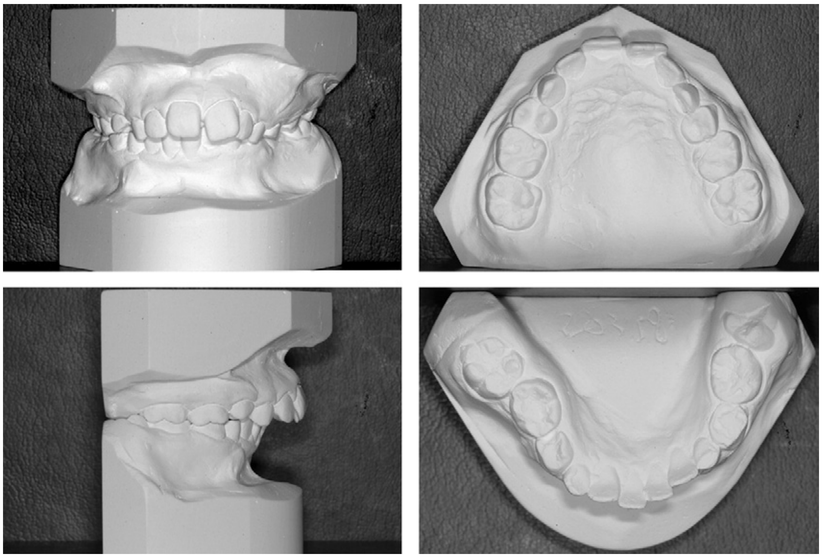

**Fig 4.** Predistraction dental models.

with the midline in the mandible. Labial gingival recession was seen at mandibular left first lower incisor. The patient was 9 years old (Figs 3 and 4).

## TREATMENT ALTERNATIVES

Several treatment alternatives were explored. However, lengthening the mandibular ramus with conventional orthognatic surgery was a poor option because it would mean waiting until growth had ceased at about 17 years of age. Furthermore, dental compensation could not be achieved because the sagittal difference between the jaws was too large, and this would not improve the airway space. Therefore, DO was the best option for mandibular lengthening.

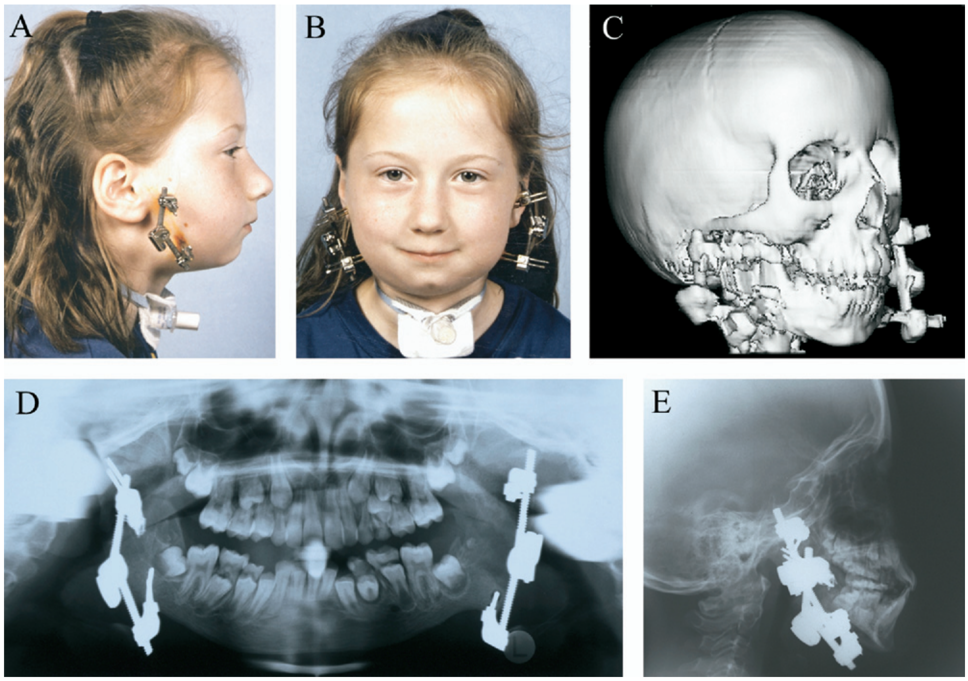


**Fig 5.** Prestabilization records at 9 years 7 months: **A** and **B,** facial photographs; **C,** 3-dimensional computed tomography scan; **D,** panoramic radiograph; **E,** cephalometric radiograph.

## TREATMENT PROGRESS

When the patient was 9 years 7 months old, an extraoral distractor (Leibirger Multiguide; Stryker, Duisburg, Germany) was surgically placed under general anesthesia. Ramus DO in the mandible was activated 9 days after the operation (usually distraction would start after 6-7 days), and the activation rate was 1 mm a day. After 13 days of activation, a stabilization period of 103 days followed. In total, the distraction was 13 mm for the right side and 11 mm for the left side (Fig 5).

The tracheostomy was decannulated 42 days after the end of the stabilization period. The patient was then 10 years 1 month old and in the late mixed dentition. She had crowding in both jaws, overjet was 1.5 mm, and overbite was 0 mm. The maxillary midline was shifted 3.5 mm to the left compared with the mandibular midline. The molar relationship was Class I (Class II tendency) on the right side and Class III on the left side (Figs 6 and 7).

An activator was placed for retention at age 10 years 11 months. Three months later, a relapse resulted in a Class II tendency, which was treated with cervical headgear. The patient wore the headgear 16 hours a day for 1 year 7 months.

Fixed appliances were placed in the mandible when the patient was 11 years 11 months old and in the maxilla at 12 years 3 months; Class II elastics were used. The fixed appliances were removed when the patient was 13 years 3 months old. After that, she wore a positioner 3 times a day for 15 minutes each and at night for 5 months; a spring retainer was then placed on the mandibular anterior teeth because of a slight relapse of the crowding.

## RESULTS

The distraction was successful, and the patient could be decannulated.

From the predistraction stage to prestabilization, the length of the mandible increased 17.7 mm, Cd-Go increased 8.0 mm, and SNB angle increased 8.4° (Table, Figs 8 and 9).

When the DO device was removed, Cd-Go was stable, but the length of mandible and the SNB angle had slightly retrogressed (by −0.4 mm and −1.7°) compared with the situation just after distraction. The net result was that the length of the mandible increased 17.3 mm, Cd-Go increased 8.2 mm, SNB angle increased 6.7°, and SN-GoGn increased 2°.

At a follow-up appointment 4 years 8 months after the DO (including fixed appliance treatment), the length of the mandible was preserved and even increased by 1.0 mm (Figs 10 and 11). Overjet was 1 mm,

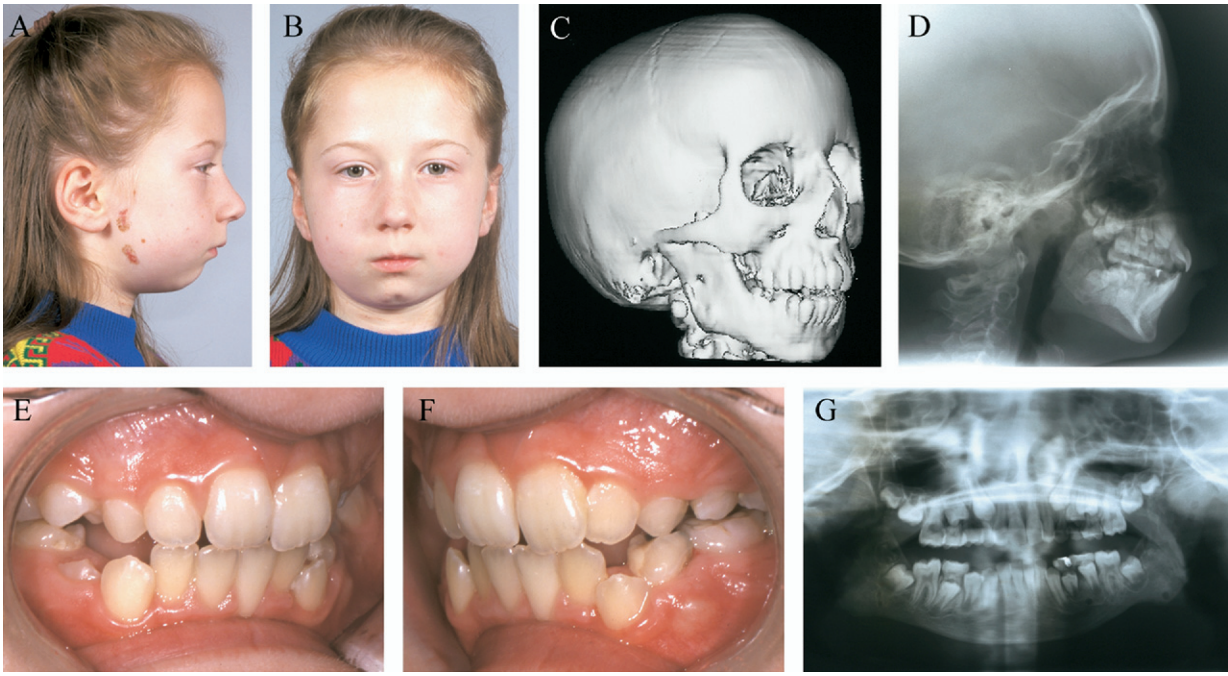


**Fig 6.** Postdistraction records at 10 years 1 month: **A** and **B,** facial photographs; **C,** 3-dimensional computed tomography scan; **D,** cephalometric radiograph; **E** and **F,** intraoral photographs; **G,** panoramic radiograph.

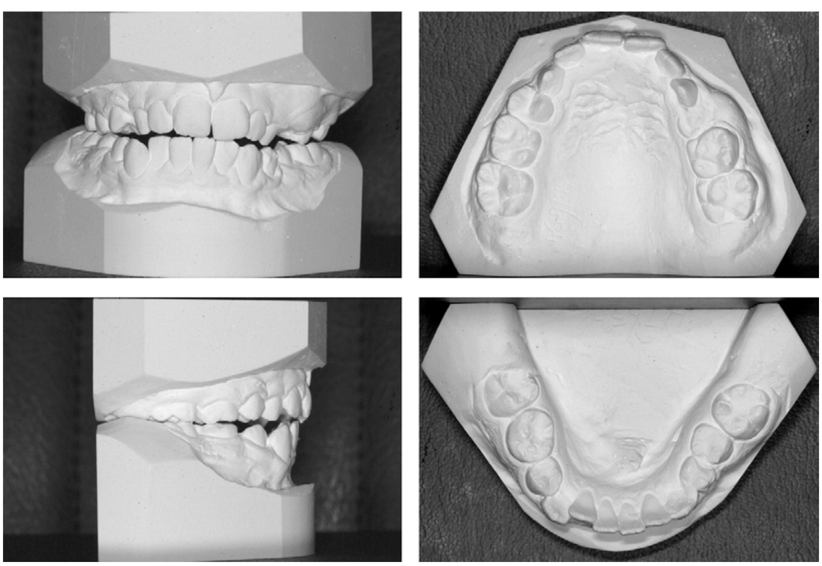

**Fig 7.** Postdistraction dental model.

overbite was 0.5 mm, and the midlines were in line. A Class I molar relationship was recorded. However, the mandibular protrusion had relapsed, SNB angle decreased by 3.1° from the postdistraction value, and the final position of chin was not optimal. A genioplasty was recommended, but the patient declined this option.

The inclination of the lower incisors to NB and to Go-Gn changed with fixed appliance treatment to

**Table.** Cephalometric norms for a 9-year-old girl and the patient's measurements during treatment and 4 years 8 months posttreatment, between brackets, measured at deciduous tooth

| *Measurement* | *Norms* | *Posttracheostomy* | *Predistraction* | *Prestabilization* | *Postdistraction* | *Follow-up* |
|---|---|---|---|---|---|---|
| SNA angle | 80.5° | 80.0° | 79.9° | 79.9° | 79.9° | 77.8° |
| SNB angle | 76.5° | 63.2° | 63.3° | 71.7° | 70.0° | 66.9° |
| ANB angle | 4.0° | 16.8° | 16.6° | 8.2° | 9.9° | 10.9° |
| Midfacial length | 88.3 mm | 66.2 mm | 69.6 mm | 72.0 mm | 72.0 mm | 79.0 mm |
| Mandibular length | 103.3 mm | 62.0 mm | 66.9 mm | 84.6 mm | 84.2 mm | 85.2 mm |
| ANS-Me (FH) | — | 39.5 mm | 40.9 mm | 50.3 mm | 49.6 mm | 48.6 mm |
| ANS-Me/N-Me | — | 49.2% | 47.2% | 51.5% | 51.1% | 48.0% |
| Cd-Go | — | 30.8 mm | 31.6 mm | 39.6 mm | 39.8 mm | 40.6 mm |
| Go-Gn | — | 38.2 mm | 43.2 mm | 51.5 mm | 51.5 mm | 52.2 mm |
| Ar-Go-Gn | 122.0° | 124.9° | 125.0° | 134.4° | 132.8° | 131.6° |
| SN-GoGn | 35.0° | 43.8° | 43.2° | — | 45.2° | 45.4° |
| Interincisal angle | 125.5° | (109.8°) | 107.2° | 123.2° | 121.1° | 112.5° |
| U1/NA | 24.8° | (8.9°) | 11.9° | 11.5° | 11.5° | 9.0° |
| L1/NB | 25.7° | 44.4° | 44.3° | 37.1° | 37.5° | 47.7° |
| L1/GoGn | 94.2° | 117.3° | 117.8° | 104.3° | 102.3° | 115.4° |

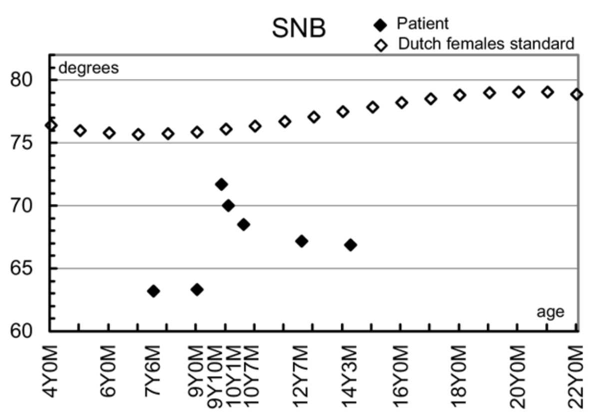


**Fig 8.** Change in SNB angle in patient compared with norms for Dutch girls.

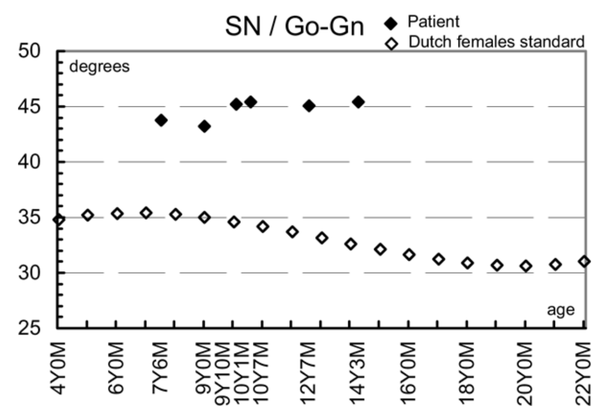


**Fig 9.** Change in SN-GoGn in patient compared with norms for Dutch girls.

compensate for skeletal relapse, even though there was a tendency toward normalization with DO treatment (Figs 6 and 10).

The girl was still small for her age (Fig 2), she did have some catch-up growth after the tracheostomy and ended up within 2 SD of Dutch female standards.

Her next check-up will be at 18 years of age. At that time, she will be dismissed if there are no problems.

## DISCUSSION

Tracheostomy can be an effective treatment for patients with severe respiratory obstruction caused by mandibular retrognathism. However, it is accompanied by high costs, frequent morbidity,[3] speech problems, and psychological problems. Therefore, it is important to investigate possible alternatives. DO has become accepted as an efficient treatment for severe mandibular hypoplasia and OSAS. The tongue base is carried anteriorly via its muscular attachments to the distracted mandible, thus pulling the tongue out of the hypopharynx and relieving the airway obstruction.[4]

This case demonstrates that DO improved this patient's airway restriction. The tracheostomy could be removed, and the patient had a more normal rate of general growth afterward (Fig 2).

After the distraction, the lengths of the mandibular ramus and the mandible increased slightly. The small increases might be explained by local bone apposition. Unfortunately, clockwise rotation of the mandible occurred during and after the DO; therefore, the skeletal Class II pattern was not improved sufficiently (Fig 12). The use of cervical headgear might have extruded the maxillary first molars. Furthermore, the forward and downward growth of the maxilla might also have had an adverse effect.

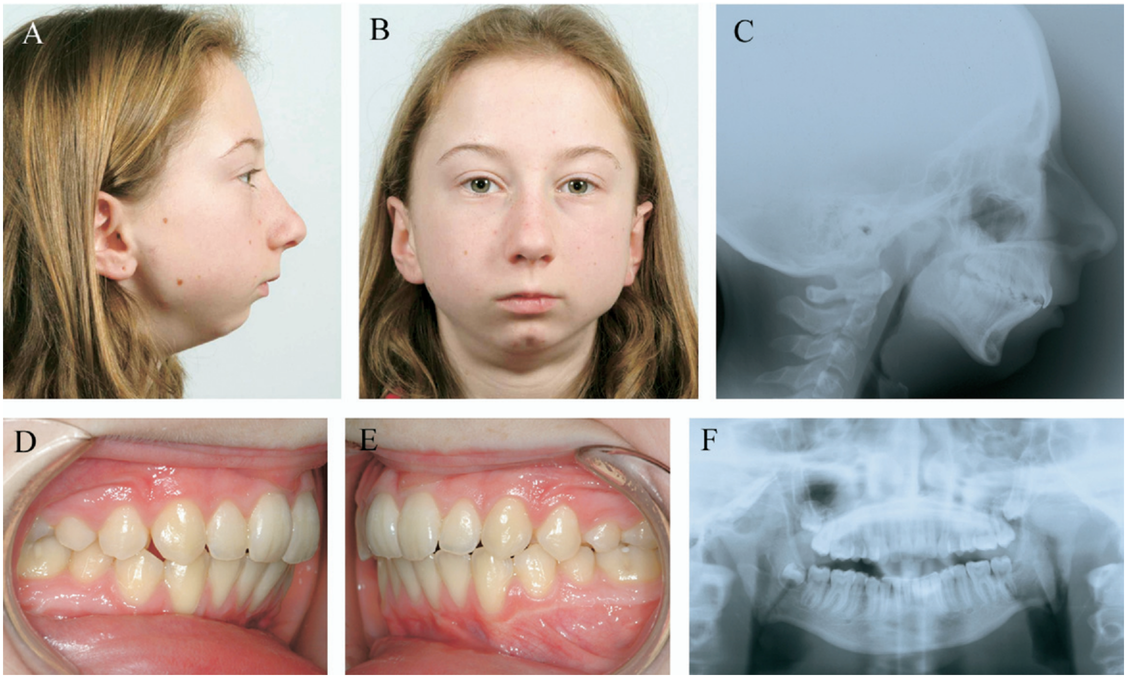


**Fig 10.** Follow-up 4 years 8 months after DO: **A** and **B,** facial photographs; **C,** cephalometric radiograph; **D** and **E,** intraoral photographs; **F,** panoramic radiograph.

**Fig 11.** Follow-up dental models.

Vertical control to prevent backward rotation of the mandible and prediction of the amount of maxillary growth and the consequent dental compensation are generally difficult to achieve, but it might be better to use vertical elastics much more radically during and after DO.

Finally, the position of the chin was not optimal. The facial profile could have been improved with chin correction, but the patient did not want further surgery.

Many studies have demonstrated the usefulness of DO for the correction of mandibular hypoplasia, and some of them described early decannulation or even

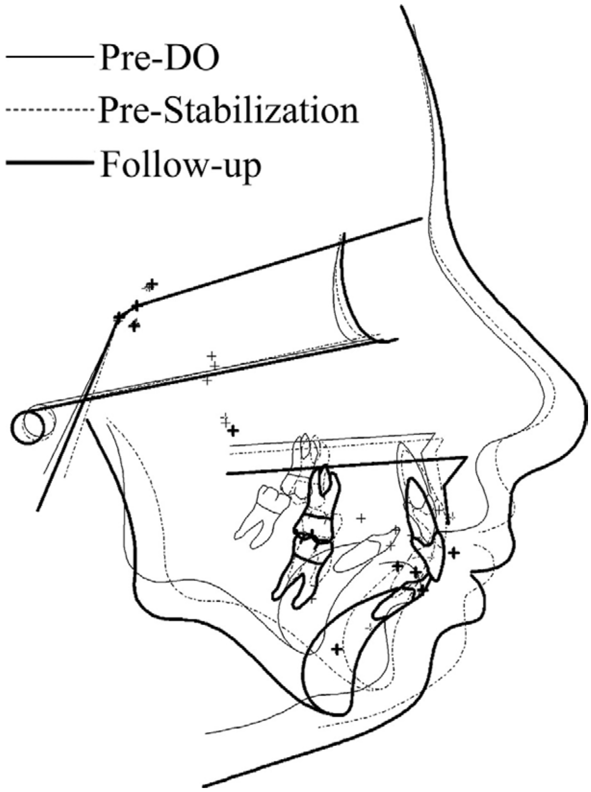


**Fig 12.** Superimposed cephalometric tracings.

avoidance of tracheostomy[5-11] in the case of OSAS. Success is claimed, but longer evaluation will be needed to prove that this treatment gives a stable result in the long term.

## CONCLUSIONS

In a patient with OSAS due to extreme mandibular hypoplasia, mandibular DO is considered useful in lengthening the mandible so that the patient obtains a normal airway condition. However, the possibility of long-term relapse can jeopardize the final result.

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