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Distraction Osteogenesis of Maxilla and Midface in Postradiotherapy Patients

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Although distraction osteogenesis (DO) is widely used, there is minimal information on its use in patients after radiotherapy. The mutilating effects of ablative head and neck surgery, and insufficient development of the craniofacial skeleton after childhood head and neck malignancies, frequently necessitate complex reconstruction techniques. The simultaneous expansion of soft tissue that comes with bony lengthening during DO is a unique phenomenon.[1] In selected cases, it causes less morbidity and better esthetic results than any other surgical procedure. The effects of radiotherapy on the outcome of DO are still not clear. There are only a few case reports describing DO of the human craniofacial skeleton after radiotherapy. Most of these reports have dealt with mandibular DO.[2-4] Only 1 case of postradiotherapy midface DO has been described thus far. Grover et al[5] presented a patient with radiation-induced orbital zygomatic hypoplasia, which was treated using a rigid external distraction device. Several animal studies have been performed to explore the advantages of hyperbaric oxygen in postradiation DO, but these studies were confined to mandibles.[6,7] In the present article, 2 patients are presented with radiation-induced midfacial hypoplasia after childhood malignancies. These patients were successfully treated with rigid external DO in combination with hyperbaric oxygen (HBO) therapy.

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Report of Cases

CASE 1

A 20-year-old female patient had cranial and midfacial hypoplasia because of radiotherapy after a bilateral retinoblastoma (Figs 1, 2). She underwent fractional radiotherapy of 45 Gy in the right and left retinas at 4 months of age, for which a D-shaped field in the craniocaudal direction was used. Because of the radiotherapy, she developed oligodontia and insufficient growth of the midface. Initially, watchful waiting was conducted. At 15 years of age, orthodontic alignment of the remaining teeth was initiated. At 20 years, she was scheduled for midface distraction. Planning was performed using stereolithography. Temporal hollowing and hypoplasia of the orbits, zygomatic complexes, and the maxilla were diagnosed. One of the main problems was the content of the orbits, which preferably would be left unaltered. The treatment plan included DO of the maxilla to restore occlusion and to correct the midfacial hypoplasia and augmentation of the temporal regions for cosmetic reasons. The patient received 20 preoperative sessions of HBO therapy, after which a standard Le Fort I maxillary osteotomy until downfracture was performed. A Rigid External Distractor (RED) frame (KLS Martin, Tuttlingen, Germany) was positioned using 2 paranasal miniplates and 2 stainless-steel transcutaneous wires of 0.4 mm each. After an 8-day latency period, DO was started at 1 mm once daily. Maxillary advancement was already visible after 8 days. After 24 days, active DO was stopped. According to the treatment plan, the patient was to receive another 10 sessions of HBO therapy. However, practical problems arose because the patient while wearing the RED frame did not fit into the HBO cap. Therefore, it was decided to postpone HBO therapy and to remove the RED frame earlier. Two months after active DO, the RED frame was removed. A Delaire protraction face mask was constructed to ensure further consolidation in a steady position during HBO therapy. Total advancement of the maxilla as measured on radiographs was 20 mm. By clinical and radiologic analyses, good consolidation was observed. Ten months after the DO procedure, the temporal regions were augmented with osteoconductive hydroxyapatite bone cement (BoneSource; Stryker Biotech, Hopkinton, MA). The patient again received 10 postoperative sessions of HBO therapy. Dental rehabilitation included extraction of the remaining primary teeth, preimplantation augmentation of the upper and lower jaws using anterior iliac crest bone, correction of the gingival margin, and placement of 7 dental implants in the upper jaw and 3 dental implants in the lower jaw, combined with a bridge suprastructure.

CASE 2

A 19-year-old male patient with a history of a nasopharyngeal rhabdomyosarcoma from 4 years of age was treated

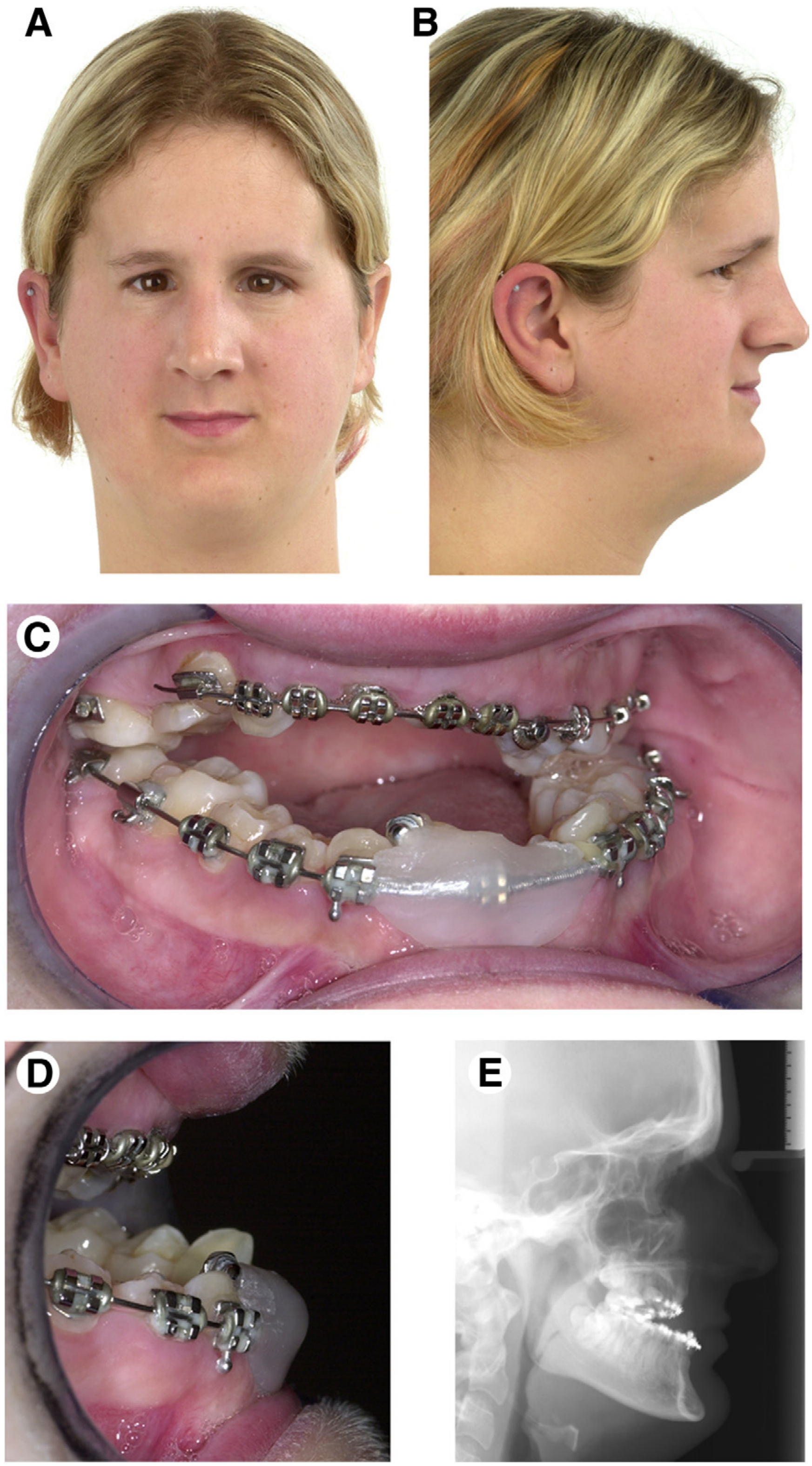

FIGURE 1. Patient 1, preoperative. *A,* Frontal view. *B,* Profile. *C, D,* Intraoral views showing a large inverse sagittal overbite. *E,* Lateral cephalometric radiograph showing midfacial hypoplasia.

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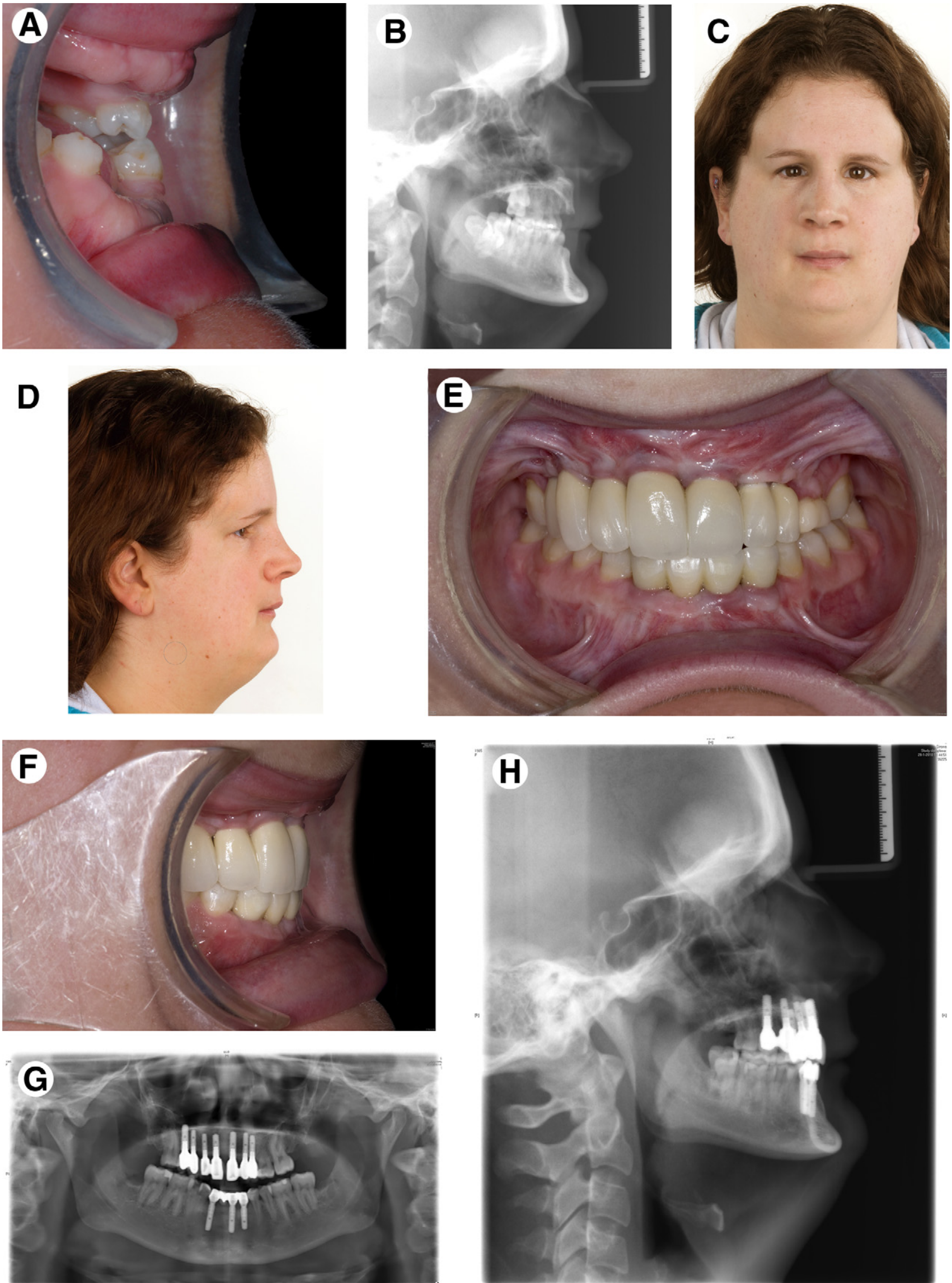

FIGURE 2. Patient 1, postoperative. *A,* Intraoral view. *B,* Lateral cephalometric radiograph showing maxillary advancement. *C-H,* Final situation 4 years postoperatively.

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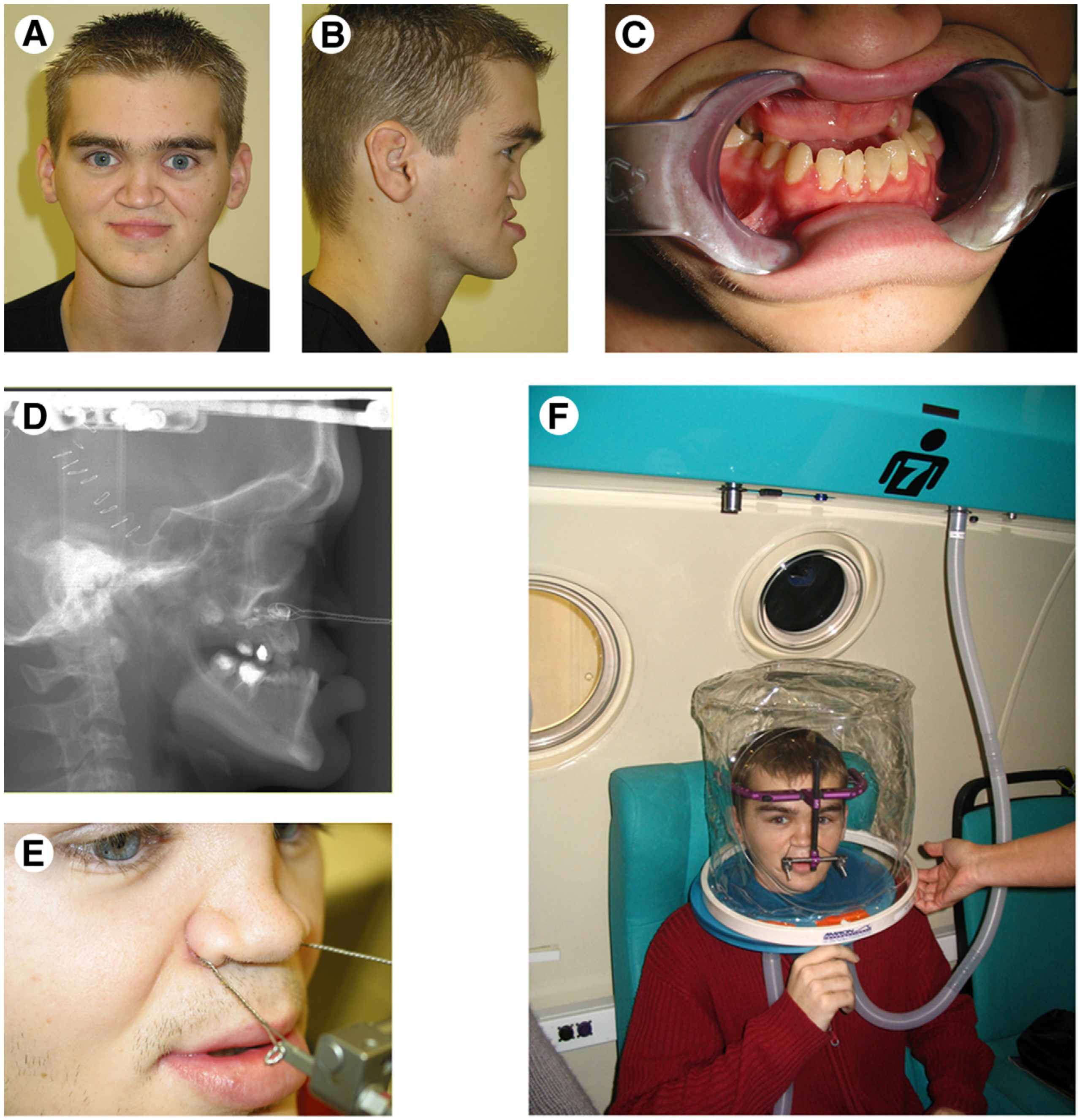

FIGURE 3. Patient 2, preoperative. *A*, Frontal view. *B*, Profile showing severe midfacial hypoplasia. *C*, Intraoral view. *D*, Lateral cephalometric radiograph. *E*, Extraoral view showing transcutaneous wires. *F*, Hyperbaric oxygen cap in situ.

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by debulking of the tumor (Figs 3, 4). Thereafter, he received chemotherapy for 10 months and underwent additional radiotherapy as local brachytherapy (11 × 4 Gy, 1 week). He displayed severe midfacial hypoplasia, insufficient dental development, and aplasia of the soft and hard palates. Because of severe underdevelopment of the dental roots, orthodontic treatment was contraindicated. Watchful waiting was conducted.

The staged surgical treatment plan was as follows. The first stage involved a Le Fort III osteotomy with extraoral DO using a RED frame in combination with 20 preoperative and 10 postoperative sessions of HBO therapy. In the second stage, reconstruction of the palate with a pedicled temporalis muscle flap and removal of the maxillary dentition was scheduled. In the third stage, implantology and prosthetic rehabilitation were adopted.

At 19 years of age, the first surgery, a standard Le Fort III osteotomy using a coronal and transmucosal approach with mobilization, was performed. The maxillary complex was connected to a RED frame using bilateral transcutaneous wires (0.6 mm) that were fixed to miniplates at the piriform aperture. After a 10-day latency period, active DO was

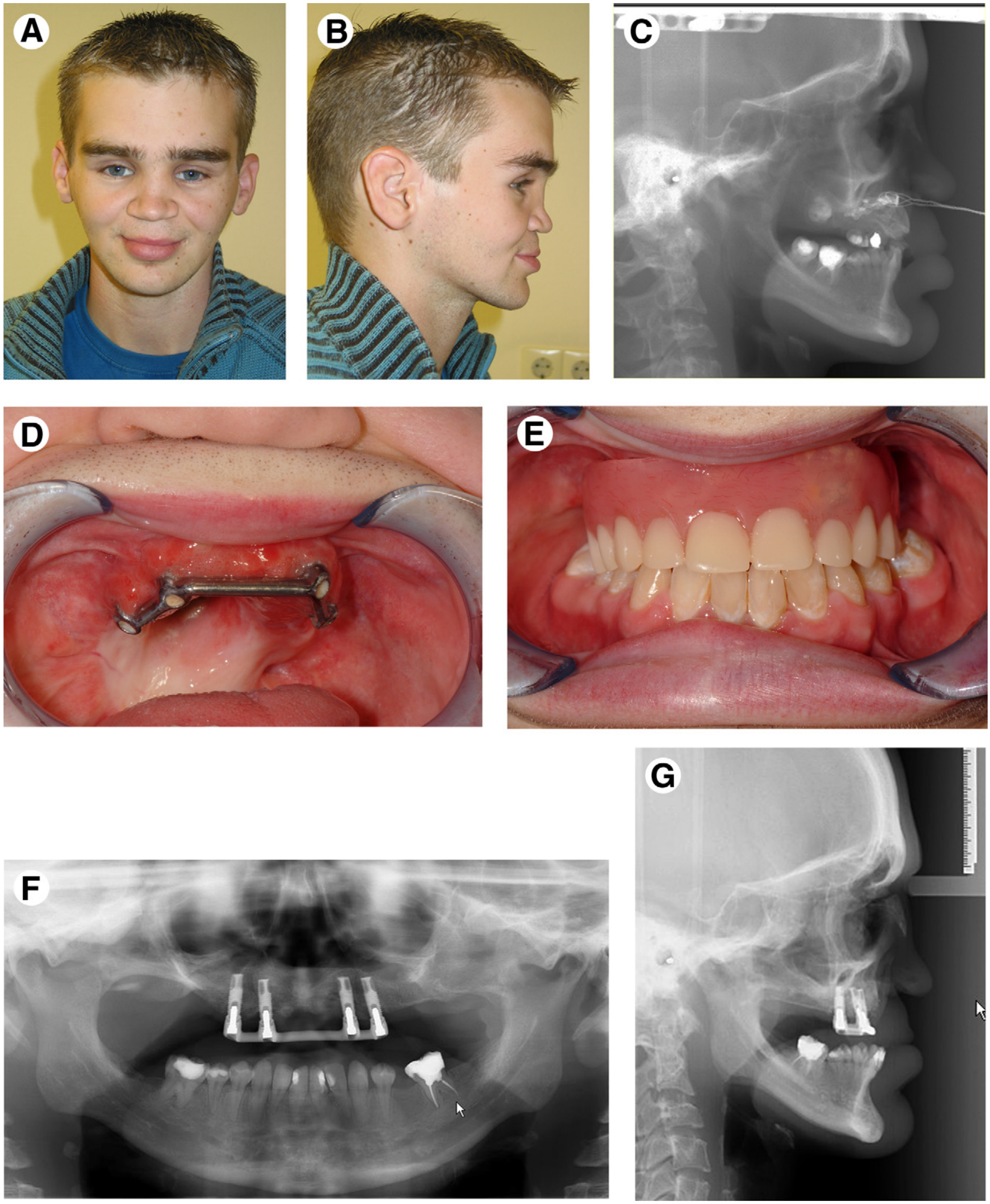

FIGURE 4. Patient 2, postoperative. *A,* Frontal view. *B,* Profile. *C,* Lateral cephalometric radiograph directly postoperatively. *D-G,* Final situation 4 years postoperatively.

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started at 0.5 mm once daily. The vector of DO was parallel to the Frankfort horizontal plane. Because of little progression after 11 days, it was decided to administer DO at 0.5 mm twice a day. After 25 days, active DO was stopped and total advancement measured on radiographs at the central incisors was 15 mm. Four months later, reconstruction of the palate was performed. A bilateral coronoidectomy was performed to decrease trismus. The miniplates on the maxilla were removed, as were the remaining maxillary teeth. The palatal defect was closed with a full-thickness temporalis muscle flap and a small caudally based pharyngeal flap. The patient received 4 dental implants in the maxilla and an implant-retained overdenture.

Discussion

This report describes 2 successful cases of midface distraction after radiotherapy with perioperative administration of HBO. DO in an irradiated midface is rarely reported, and the parameters of DO are empirically used in these cases.

Clinical parameters that affect treatment outcomes of craniofacial DO include age, surgical technique, distraction rate, latency period, consolidation period, and type of distraction device.[1]

In nonirradiated patients, the optimal rate of distraction is 1 mm/d. Faster rates have been reported to be successful in midface distraction.[1,8]

In patients after irradiation, different rates and different outcomes have been reported, but the optimum values remain unclear.[2-5,9] The findings in the present case study showed a successful 1-mm rate for midface distraction after radiotherapy.

A latency period is necessary for initial callus formation and soft tissue healing. The initial callus responds to tensile stress created during distraction.[1,6] A 5- to 7-day latency period for mandibular DO and a 4- to 5-day latency period for maxillary DO are the most common.[1] In midface DO, latency periods vary from 5 to 7 days to starting DO immediately.[1,9] In irradiated tissue, because of cellular loss and hypovascularity, callus formation and bone healing presumably take more time. A longer latency period should therefore be considered.[6]

Taub et al,[9] however, started DO of an irradiated palate after 2 days, with good bone formation. In this postradiotherapy midface DO case study, a latency period of 8 to 10 days produced good results.

A recommended consolidation period of 2 to 3 months has been reported for maxillary and for midfacial DO. A longer consolidation period for the latter has been suggested because of the thin structure of the bone at the distraction sites.[1]

External DO in a case with radiation-induced orbital zygomatic hypoplasia produced satisfying bone formation but not at all osteotomy sites.[5] DO was administered for 7 months on a variable schedule. It was suggested that the vector of distraction is an important factor. If various vectors are used subsequently, stress on the callus could come closer to compression than to tension, thereby inhibiting bone formation.[5] In the 2 patients in the present study, 1 vector was used with only small adaptations, and a 3-month consolidation period was taken into account, after which good clinical bone formation was found.

Other factors that might affect DO treatment outcome could be the interval between radiotherapy and DO, previous extensive ablative procedures, or a radiation dose threshold. In the literature, radiation-to-distraction time has varied from 18 months to 20 years. The negative effects of radiotherapy for childhood malignancies usually are not immediately visible, but become clear during the growth period, causing hypoplasia of the affected area. Unsuccessful bone formation has been reported in patients with short postradiotherapy intervals, and successful bone formation has been shown in patients with longer intervals.[2,4,5,9] Although cellular loss and hypovascularity after radiotherapy worsen over time,[10] the extended time from radiation to distraction in these young patients appears to be a positive factor for treatment success. In this study, the intervals were 20 and 15 years, respectively.

Apart from radiation, previous ablative procedures may impair the quality of the tissues. One case report described an 80-year-old patient who had undergone many oncologic surgical procedures before DO was performed in the mandible; the DO failed.[4]

There might be a radiation dose threshold above which DO becomes troublesome.[4] In 2 reported cases of mandibular DO, a cumulative dose of 60 Gy to the later distraction site was administered. In these 2 patients, new bone formation failed.[3,4] Two cases of successful DO, however, were reported after radiation dosages of 45 Gy and 60 to 70 Gy, respectively.[2,9] In the present case study, the patients received 90 Gy (2×45 Gy) and 44 Gy (11×4 Gy, brachytherapy), respectively. Both showed satisfactory and stable bone formation. According to these results, no threshold for radiation can be defined. Apparently, DO can be successful even after radiation dosages higher than 60 Gy. Of course, differences in vascularization between the mandible and midface should be taken into account.

Although extensive studies have been conducted on the use of HBO, there is still no consensus on its value. HBO is thought to improve healing of radiation-damaged bone by increasing angiogenesis and is believed to change the pattern of bone-forming activity toward that of nonirradiated bone.[6,7,10] In the available case reports on DO after radiotherapy, some investigators have reported satisfactory bone formation without using HBO,[2,5,9] whereas others have reported failures while using it.[3,4] The 2 present patients received pre- and postoperative HBO therapy to decrease the risks of nonunion and osteoradionecrosis. Postoperative problems arose because it was not possible to fit the heads of the patients with the RED frame on into the HBO cap without making technical adjustments. Distraction of the midface was successful using HBO in these cases, but this has only anecdotal relevance.

In addition to the published parameters that influence the treatment outcome of DO, use of transcutaneous fixation instead of transoral fixation might contribute to the successful treatment outcome.

From this case study, it can be concluded that midfacial DO in combination with HBO therapy can be a safe and successful therapy to treat developmental disturbances in patients with midface hypoplasia after radiotherapy. The functional and esthetic results are satisfying. This approach can be considered when classic osteotomies of the midface are thought to coincide with higher morbidity and less stability. Parameters influencing the success might be DO confined to the midface, perioperative HBO therapy, and use of transcutaneous wires.

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