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Treatment of a Patient with Cleidocranial Dysplasia Using a Single-Stage Implant Protocol

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Abstract

This patient report describes the treatment of a 45-year-old Caucasian woman with cleidocranial dysplasia who had significant dental problems that greatly affected her quality of life. The patient had orthodontic treatment in her earlier years along with surgical removal of supernumerary teeth. Using implants, the maxillary and mandibular arches were restored with fixed screw-retained prostheses. Eight implants and six implants were placed in the maxilla and mandible, respectively. Both arches were immediately loaded following the Teeth in a Day™ protocol using an all-acrylic resin provisional prosthesis. Five months later, definitive maxillary and mandibular prostheses were fabricated. The patient has been followed for a period of 5 years, and all postoperative evaluations have been uneventful.

In 2005, the patient described in this article had just seen her local dentist in Sydney, OH. Her dentist recommended extracting all her teeth and placing maxillary and mandibular complete dentures. The patient had reviewed the dental literature and come across a case study the authors of this report had published where a patient with her similar congenital condition was successfully treated using dental implants. She immediately contacted the treatment center asking if there was any hope for her. In response, she was told not to have any of her teeth removed and that she could be helped if she could travel to Pennsylvania. This article describes the treatment that significantly enhanced her quality of life.

In 1897, Marie and Sainton[1] first described cleidocranial dystosis, a rare inherited skeletal dysplasia. It has since been known as cleidocranial dysplasia. Its underlying pathology is a generalized skeletal dysplastic condition.[2] It is an autosomal dominant pattern of inheritance; however, it has been suggested that between 20% and 40% of cases represent new mutations.[3] It is best known for its dental and clavicular abnormalities, and it is a disorder of bone caused by a defect in the CBFA 1 gene of chromosome 6p21. This gene guides osteoblastic differentiation and appropriate bone formation when expressed under normal conditions.

Individuals with cleidocranial dysplasia tend to be of short stature and have proportionally large heads with pronounced parietal and frontal bossing. A broad base of the nose with a depressed nasal bridge as well as ocular hypertelorism can be observed. The most dramatic finding odontologically is the

presence of numerous unerupted permanent and supernumerary teeth.

The dental abnormalities associated with cleidocranial dysplasia represent a remarkable dental challenge.[4] Treatment of the dental problems associated with cleidocranial dysplasia may be difficult.[4] These treatments are difficult due to the supernumerary teeth, malformed and shortened roots, delayed eruption of permanent teeth, and underdeveloped maxilla and mandible. The treatment often involves multidisciplinary approaches with a combination of orthodontics, orthognathic surgical interventions, and interim prostheses. This process of involving all these disciplines can take several years until patients can receive their definitive prostheses.

Therapeutic dental options include a crown sleeve coping overdenture,[5] full-mouth extractions followed by denture fabrication, autotransplantation of selected impacted teeth followed by prosthetic restoration, or removal of primary and supernumerary teeth followed by exposure of permanent teeth subsequently extruded orthodontically. The use of implants in a patient with cleidocranial dysplasia has been documented to support a removable implant-retained overdenture.[6] Petropoulos et al[7] documented a treatment using implants to support an implant-supported fixed prosthesis. To date, there is a limited amount of documented cleidocranial dysplasia treatments using an implant-supported fixed prosthesis with immediate loading. Using implants in such situations seems logical, since there have been documented cases of bone formation around teeth that have been orthodontically erupted in patients with cleidocranial dysplasia.

Clinical report

Patient history

The patient was a 45-year-old woman (Figs 1A–C) born with cleidocranial dysplasia. She was missing a piece of her clavicle (Fig 1D) and had other facial anomalies common with this condition. Her father, middle sister, daughter, and son also carry this genetic defect. The patient was in excellent general health, with allergy to codeine, when presenting for treatment related to reconstruction of her dentition (Fig 1E). Her past dental history revealed that at age 12, she had undergone multiple tooth extractions (about 30 teeth), which took 6 hours (according to her recollection), where all her deciduous teeth and supernumerary teeth were removed (Figs 2A–C). Subsequently, she underwent orthodontic treatment for 7 years. Following these treatments, she was strongly dissatisfied with her appearance. This strong discontent continued throughout her life. Her teeth were misaligned and did not show when she spoke and smiled. Additionally, some of her teeth were loose. Her dentist told her to be satisfied with what she had, as she would "never have that Colgate smile." She was advised to have all her teeth extracted; however, she was afraid and concerned about not being able to function properly with removable dentures. She was also told that due to excessive bone loss she was not a candidate for dental implants. Her chief complaints were, "I have some missing teeth," "I am unable to chew properly," "my teeth do not show when I speak," and "I am very ashamed of my appearance and extremely dissatisfied" (Fig 1B).

Clinical evaluation and diagnosis

At the initial visit, the patient presented with the following teeth in the maxillary arch: #s 1–6, 8, 9, and 11–16 (Fig 2C); and in the mandibular arch: teeth #s 18–20, 22–25, and 27–31. Teeth #17 and 32 were unerupted and had two supernumerary teeth in the posterior maxilla and one in the mandible.

Clinical examination revealed that the patient was in a dental class I relationship with an anterior cross bite and malaligned teeth. She showed severe occlusal wear on her posterior teeth, loss of occlusal vertical dimension (OVD, Fig 1C), and severe horizontal and vertical bone loss. She had generalized chronic advanced periodontitis. Tooth #6 was abscessed and extremely loose and was extracted prior to fabricating diagnostic casts. Additionally the patient had periodontal abscesses around #8 and 13. She was classified according to the ACP Prosthodontic Diagnostic Index classification for partial edentulism as Class IV.[8]

Diagnostic casts were made and articulated at her existing OVD (Fig 2D). Intraoral and extraoral visual examination determined that the patient had an existing interocclusal rest space of 7 mm (Fig 1C). A second set of diagnostic casts were made and articulated at a newly reestablished OVD, giving the patient 3 mm of freeway space. The reestablished vertical dimension was recorded with an interocclusal bite registration (Regisil, Denstsply York, PA). Diagnostic casts were articulated at this improved and increased OVD, which was later used to fabricate transitional immediate dentures for the maxillary and mandibular arches.

The patient had short conical roots along with generalized chronic advanced periodontitis, which did not make her a good candidate for orthodontic intervention. Her remaining dentition had a poor prognosis to serve as abutments for fixed restorations. Other options included complete maxillary and mandibular dentures, or implant-retained overdentures. A complete maxillary denture would have presented retention challenges for the patient due to her maxillary palate being extremely shallow; she also had a severe gag reflex. Not replacing the teeth with implants would also have allowed continued ridge resorption in both arches.[9] The patient expressed her desire for something not removable and desired fixed prostheses. She was presented with the fixed implant reconstruction and accepted this treatment approach.

Clinical treatment

Local anesthesia was administered using Marcaine (Abbott Laboratories, Abbott Park, IL) 1:200 × 5 carpules and Lignospan (Septodent, New Castle, DE) 1:50 × 5 carpules. All maxillary and mandibular teeth were extracted. Crestal incisions were made, along with flap elevation for both arches. The necessary anatomic landmarks in both arches were identified prior to making any preparations in the bone. In the mandibular arch, the critical anatomic landmarks are the mental foramina, positioning of the inferior alveolar canal, and positioning of the anterior loop; these are identified visually. The anterior loop was carefully probed to determine the extent of the anterior extension. In the maxilla, the floor of the nose could be visualized from a labial flap elevation. Additionally, the floor and the

Figure 1 (A) Patient at 2 years old. (B) Preoperative full face showing a broad base of the nose with a depressed nasal bridge. (C) Concave preoperative profile view of patient showing loss of vertical dimension. (D) Approximation of clavicles; characteristic of cleidocranial dysplasia. (E) Preoperative intraoral photograph of maxillary and mandibular teeth in occlusion.

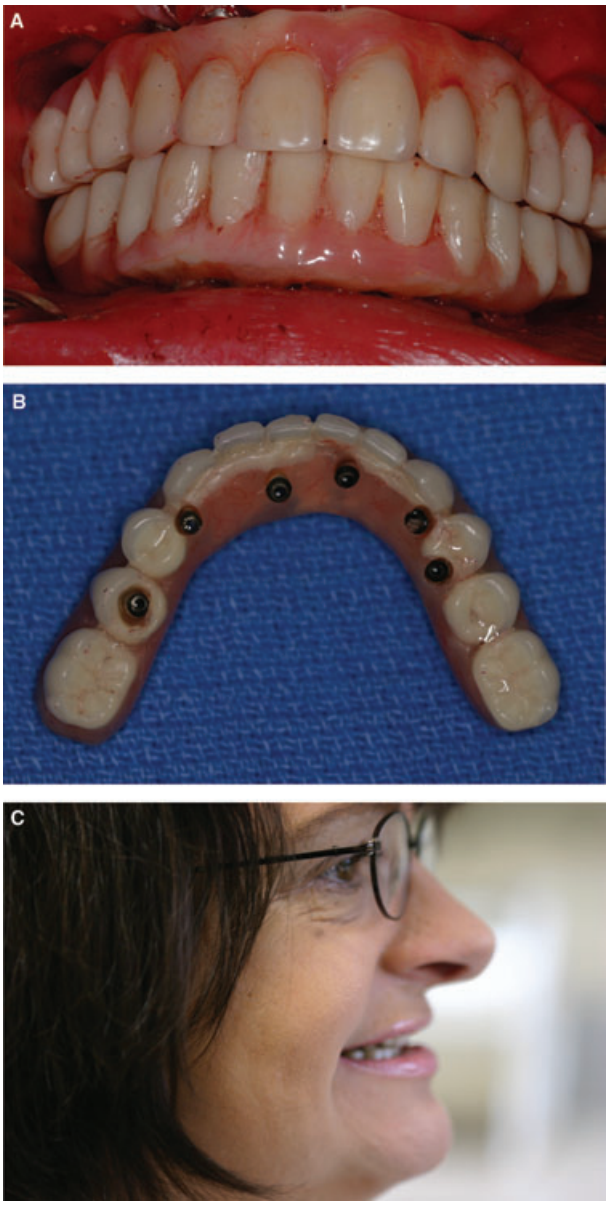

Figure 3 (A) Maxillary and mandibular conversion prostheses. (B) Mandibular conversion prosthesis. (C) Profile view of maxillary and mandibular conversion prostheses in place.

walls of the sinuses could be visualized through the thin lateral walls.

Prior to surgery, the determination on the position of the implants and the decision of whether to bone graft were made from panoramic radiographic analysis. The patient and the clinicians were ready to revert to a two-stage procedure if inadequate bone was available for implant placement following the removal and debridement of the alveolar bone. This would obligate the patient to wear interim complete removable dentures during the time of bone healing. Fortunately for this patient, following the removal of all her teeth, adequate bone remained for implant placement.

The implants were placed without a surgical guide. Freehand surgery was done with fully elevated mucosal flaps to allow optimal visibility of the bone. Saline irrigation was used throughout the implant drilling and placement procedure. Table 1 shows the distribution of the 14 implants used to support this patient's prostheses. The implants were coated with autogenous platelet

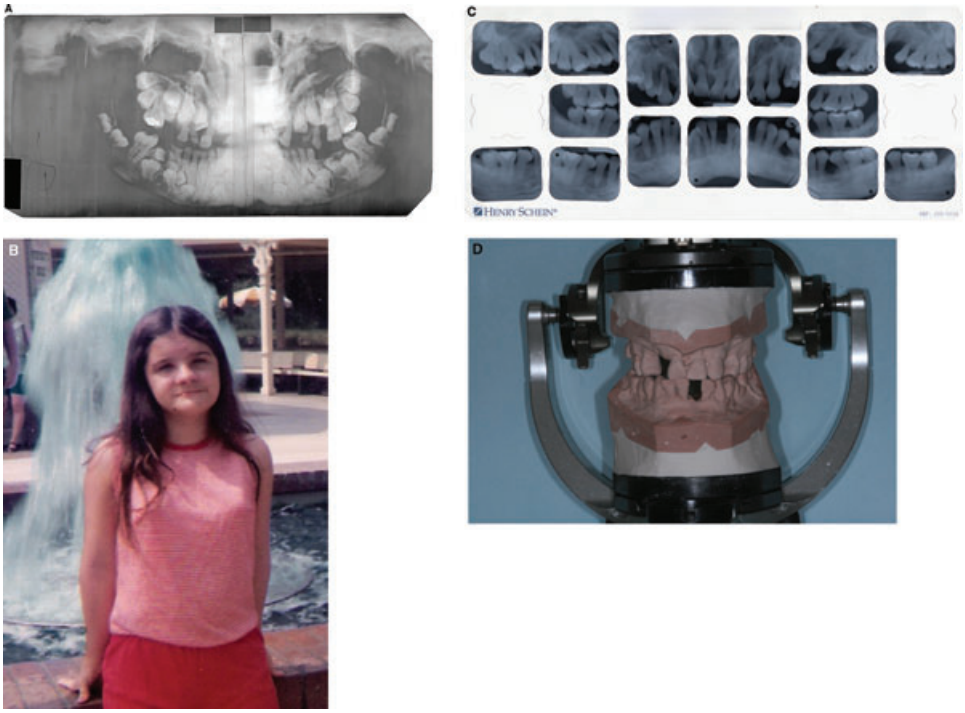

Figure 2 (A) Panoramic radiograph showing numerous supernumerary and deciduous teeth taken in 1971 when patient was 11 years old. (B) Photograph of patient at 13 years old. (C) Full-mouth series radiographs taken in 2005 when patient was 45 years old. (D) Pretreatment articulated mounted diagnostic cast.

Figure 4 (A) Maxillary metal-ceramic prosthesis at bisque bake stage. (B) Postoperative esthetics of final prostheses. (C) Postoperative esthetics. (D) Profile view esthetics.

Table 1 Location of implant (tooth #), implant size, and type

Location of implant (tooth #)	Implant size and type
3,4,7	13 × 4 mm MKIV
6	15 × 4 mm MKII
9,11	13 × 3.75 mm MKIII
13,14	13 × 4 mm MKIII
20	13 × 3.75 mm MKIII
22,24,25,27	15 × 3.75 mm MKIII
29	15 × 4 mm MKIV

rich plasma (PRP)[10] and placed using a torque-controlled machine (45 Ncm) and checked manually. Primary closure of the incisions was made with interrupted Vicryl 4.0 sutures (Ethicon Inc., Somerville, NJ).

The Teeth in a Day™ protocol[11] was followed by placing transmucosal abutments (Esthetic one and Standard, Nobel Biocare, Yorba Linda, CA) on all the Brånemark System implants. A rubber dam was then used to isolate the abutments from the mucosa and underlying bone. The prosthetic components were installed on the abutments with moderate length guide pins. All the maxillary and mandibular implants were immediately loaded using the interim complete denture prostheses made from the diagnostic casts, which were previously articulated at the newly reestablished OVD, and were converted to all-acrylic resin conversion prostheses by connecting the interim complete denture prostheses with the prosthetic cylinders using autopolymerizing resin (Jet; Lang Dental Manufacturers, Wheeling, IL). The conversion prosthesis[12,13] was first described at the 1986 International Congress on Tissue Integration in Oral and Maxillofacial Reconstruction in Brussels, Belgium. The implant-supported all-acrylic resin conversion prostheses were delivered (Fig 3). The newly established OVD was checked using phonetic tests, checking the lip support, and extraoral measurements. The patient had 3 mm of interocclusal rest space.

Postsurgical patient management

Following the mandibular and maxillary implant surgeries, the patient was provided with postsurgical instructions, including cold therapy, standard medications of Pen VK 500 mg (SmithKline Beecham Corp., King of Prussia, PA), Peridex (Dentsply Caulk, Milford DE), and Decadron 0.75 mg (Merck and Co., Inc., Whitehouse Station, NJ), and a soft diet for 8 weeks.

Definitive prostheses for maxillary and mandibular arches

Five months following surgical and provisional restorative procedures, the patient presented for fabrication of the definitive prostheses for the maxillary and mandibular arches. An interocclusal registration was made using Regisil. The final impressions were made using the existing maxillary and mandibular fixed all-acrylic resin conversion prostheses as an impression splint. Heavy body Reprosil (Dentsply) impression material was syringed beneath the prostheses, and pick-up impressions were made using long guide pins (Nobel Biocare). The master casts were created by placing abutment analogs into the temporary cylinders within the all-acrylic resin prostheses. Additionally, maxillomandibular relation records were made using the conversion prostheses at the tested OVD. Using the immediately loaded conversion prosthesis as an impression splint has been shown to create an exceptionally accurate master cast. The laboratory then began fabrication of the definitive metal-reinforced implant-supported prostheses. The patient was appointed for a variety of try-ins, including functional and esthetic assessments as well as verification of the recorded OVD.

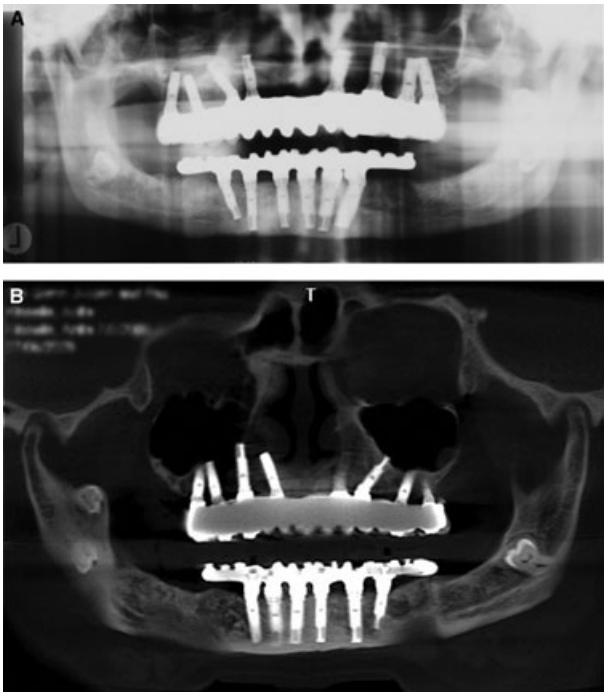

Figure 5 (A) Panoramic radiograph following definitive prosthesis delivery. (B) Follow-up panoramic radiograph 5 years postoperatively.

A metal-ceramic prosthesis was planned for the maxilla, and a gold framework wrapped with acrylic was planned for the mandible. The gold casting frameworks were tried in 2 weeks following the final impressions. The fit was verified clinically using the single-screw test and also reconfirmed radiographically.[14] At final delivery, the occlusion was adjusted so that all the contacts were even. The access holes were sealed using cotton and Fermit (Ivoclar Vivadent, Buffalo, NY). The patient was extremely pleased with the results (Fig 4). A maxillary occlusal guard was made to be worn at night. The patient has been followed on a yearly basis for maintenance visits (Fig 5) for the past 5 years. She resides in Sydney, OH, and is being seen by a local dentist.

Discussion

Brånemark System implants were used successfully in treating a 45-year-old woman with cleidocranial dysplasia. The patient

had struggled odontologically from a combination of delayed eruption, malformation, and the absence of many of her permanent teeth. The patient had undergone years of orthodontic treatment along with the extraction of her supernumerary and deciduous teeth. Due to her current state of generalized chronic advanced periodontitis, along with short conical roots, her remaining teeth were unable to serve as useful abutments for a fixed reconstruction, and it was necessary to edentulate the patient, followed by immediate implant reconstruction. The removal of all her teeth without this immediate reconstruction would most likely have left the patient with an edentulous state that would continue to deteriorate due to ongoing alveolar atrophy. The treatment described in this report provided the patient with an excellent long-term prognosis and had a huge positive emotional impact. Following her treatment, she stated that her "impossible dream of having teeth became possible." Moreover, her self-confidence grew to the point that she became an enthusiast for fellow patients with similar afflictions, starting a blog and a support group.

Summary

Using implants to support fixed prostheses is an effective treatment option for patients with cleidocranial dysplasia and eliminates the long-standing struggle with ill-fitting, uncomfortable, or unsightly removable prostheses. The Teeth in a Day™ protocol far exceeded the patient's expectations for clinical rehabilitation. The patient's care was executed by a team consisting of surgical prosthodontists, a biomedical engineer, laboratory technicians, and support staff focusing on the patient's emotional well-being. Her dental makeover took only 1 day. This aspect of her treatment cannot be overstated. Final prosthesis delivery occurred in less than 5 months. This treatment protocol is an improved standard of care for patients suffering from cleidocranial dysplasia.

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