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Full mouth implants rehabilitation of a patient with ectodermal dysplasia after 3-Ds ridge augmentation. A clinical report

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Abstract
A 22-year-old female presented with hypodontia and severe atrophy of alveolar ridge associated with ectodermal dysplasia, was treated with 3Ds bone augmentation technique and full mouth implants rehabilitation. Bone blocks harvested from the retro molar area to construct adequate bone volume at posterior maxilla and mandible. The vertical and horizontal bone augmentation allowed proper implant placement according to prosthetically-driven implant placement concept. Ten implants were placed. Six in the maxilla and four implants were inserted in the mandible. A cement-retained full mouth metal ceramic FPDs were fabricated. The outcome of the treatment showed that dental implants placed in bone block grafts should be considered as a good treatment modality for patients with ectodermal dysplasia. It also, provided the patient with a prosthesis that enhanced the patient function and esthetics.

1. Introduction

Ectodermal dysplasia (ED) comprises a large heterogeneous group of inherited disorders that are characterized by primary defects during the development of two or more tissues that are derived from embryonic ectoderm. The tissues primarily involved are the skin, hair, nails sweat glands and teeth [1]. Oral findings often are important and can include multiple abnormalities of the dentition (such as Anodontia, hypodontia, or malformed peg like teeth), loss of occlusal vertical dimension, and protuberant lips. Also, patients with ED, because of tooth absence, have hypoplastic alveolar bone with knife-edge morphology result in bite collapse, making implant reconstruction a challenge [2–4].

Advanced alveolar bone loss (>7 mm) may result in esthetically and functionally compromised dental prosthesis like removable and fixed partial dentures and ideal implant placement in prosthetically driven position [5]. The end goal of the therapy is to provide a functional restoration that is in harmony with the adjacent natural dentition. Thus augmentation of bone is often necessary [6]. Advances in biologic understanding of different bone regenerating materials and continuous innovations in surgical techniques have led to increased predictability in reconstruction of alveolar ridge defects and functional implant placement [7].
Augmentation of insufficient bone volume can be brought about by different methods, including particulate and block grafting materials, Guided Bone Regeneration with or without growth and differentiation factors, ridge splitting, expansion and distraction osteogenesis, either alone or in combination. These techniques may be used for horizontal/vertical ridge augmentation, socket preservation and sinus augmentation [6].

In an alveolar ridge with insufficient height or width to accommodate an implant with the desired dimensions, a two-stage procedure is indicated. These three-dimensional crestal defects showed unpredictable results when reconstructed with bone substitutes. Hence, reconstruction of large defects requires horizontal and/or vertical augmentation of autogenous bone grafts [8,9]. Therefore, particulate autogenous bone or autogenous bone blocks in combination with resorbable or non-resorbable membranes can be used [10]. Alternatively, a vertical augmentation can be done in combination with distraction osteogenesis, sandwich or interpositional techniques [11]. However, additional bone grafting at the time of the re-entry may be needed [12–14].

2. Case presentation

A healthy 22-year-old female dental student was presented seeking prosthetic rehabilitation. On clinical intraoral examination, the patient has suffered from ectodermal dysplasia ED that is manifested by hypodontia (only two centrals, two laterals with small roots and two stunted molar in the maxilla and six anterior teeth, one loose left premolar and two stunted molars in the mandible) and severe alveolar atrophy in the posterior ridges. The patient lived all her life on soft food. Deposits, gingival inflammation and yellowish stains were also detected around all remaining teeth. Moderate class III maxilla–mandibular relation was evident. Severe ridge atrophy in both maxilla and mandible was obvious (Fig. 1).

Extra oral examination revealed good normal conditions of hair, eye lashes, eye brow, nails and skin. Also, she has sunken upper lip and cheeks and protuberant lower lip. A speech defect of some letters was noticed (Figs. 2 and 3).

Questioning the family’s history, it was found that an older brother to the patient (23 years old) is free from any manifestation while a younger brother (17 years old) have the same manifestation of ED as his sister, However, he has more remaining teeth in his mouth. No family history of ED could be detected in any member of the family both in the father or mother sides.

Radiographic evaluation with Cone Beam Cross Sectional Tomography (CBCT) revealed that the ridges in both the maxilla and mandible were so atrophied horizontally and vertically with a knife edge ridge shape. Measurement indicated that the ridge was only 1.2 mm in the anterior maxilla and 4 mm in the premolar area of the mandible. The roots of the four mandibular incisors were almost absent. Distal caries was detected in the two mandibular canines and required root canal treatment (Figs. 4 and 5).

3. Setting the treatment plan

The treatment plan included building-up and grafting the atrophied ridges in 3Ds, applying a prosthetically-driven
implant placement concept to receive osseointegrated implants in the grafted areas. These implants will carry a full arch prosthesis to rehabilitate the severely atrophied jaws and restore the esthetic and functions of the patient. The treatment plan employed a staged approach and was divided into three stages. The first stage aimed to graft the atrophied ridges and in the second stage, implants were placed after maturation of the graft, and finally the prosthetic rehabilitation could be finalized.

Impressions and jaw relation records were obtained from the patient and used to mount the diagnostic casts on articulator. A prosthetically-driven implant concept was applied. Full set-up of the teeth was carried out. Accordingly, a transparent surgical implant guide was fabricated. Studying the full set-up of teeth with the CBCT and the anticipated positions of the implants, the size and the position of the bone grafts was decided.

Prior to the day of surgery, patient started on an oral regimen of steroids and antibiotics to prevent postoperative swelling and infection [18].

The patient was admitted to a hospital and surgical procedures were carried out under general anesthesia and local infiltrations. After induction of anesthesia, the surgical area was scrubbed and draped following a standard protocol.

4. Preparation of the recipient site

The recipient site to be grafted was accessed prior to beginning surgery at the donor harvest site. This sequence is important so that the recipient site was measured and assessed to determine the size, volume and shape of bone block(s) required to achieve the desired result. Use of a surgical guide which indicates the desired final implant position is helpful to assess the amount and position of grafting required. Crestal incision and full thickness flap reflection was the preferred method at the recipient site. A knife edge type ridge was seen on reflection of flap.

The recipient site was prepared first in the right side of the maxilla and then the donor site in the right side was prepared. After finishing the right side, the left side was prepared. The same procedures were carried out in the mandible.

Incisions and reflection was made wide enough for complete access to the site, and for adequate access for later tissue release for passive closure.

5. Retro molar block harvest

The incision design for access to the ramus was proceed by vestibular approach along the anterior border of the ramus running along the external oblique ridge. Ramus block graft is out-fractured with chisels via gentle tapping, the periphery of the osteotomy is redefined with the chisel and gentle out-fractured. This technique has the advantage of not disturbing the periodontium of the adjacent teeth. The thickness of the ramus block was 3 mm. The block dimensions were 2.2 cm width and 1.3 cm height (Fig. 6). The bone incision started with two stop cuts outlining the area to be harvested. The bone cuts were connected along the superior, then inferior borders.
6. Bone block preparation, fixation and closure of the recipient site

The fixation procedure is essentially the same whether the block is sourced from the ramus or the symphysis. A separate kidney dish of sterile saline is used to store the block and make adjustments with rotary burs in a straight handpiece, holding the block firmly using graft holding forceps.

Splitting the block into two halves was carried out first. Then the free bone block was first shaped and adapted to the recipient site. Care should be taken not to reduce the block extensively in order to achieve this fit, since this sacrifices critical volume of grafted bone Fig. 7.

Alternatively extensive adjustment is required; the recipient site can also be shaped, then cortical perforations were made. Once stability and intimate contact has been achieved, the fixation screw sites were selected and the block was penetrated in a “lagged” fashion with a twist drill which is larger than the final screw diameter so that the fixation screw threads will not engage the block, but rather engage only the cortical bone of the underlying recipient site. Use of at least two screws is recommended, since total immobility of the block graft during healing is a critical factor in successful bone integration [15]. The lagged block was stabilized in place at the recipient site with a large hemostat and the first screw site was drilled with the appropriately sized twist drill through the first lagged block hole. A periodontal probe was then placed in the first hole to retain position, and the second screw site prepared. The block is then removed and intra-marrow penetration of the recipient site is performed using a small diameter twist drill (1.0 mm) to optimize blood supply and new bone formation around the new graft.

The same procedures were carried out to fix a vertical and horizontal bone blocks to build-up a room with two walls Figs. 8–10.

Additionally, the block itself was penetrated to facilitate vascular ingrowth. At this time periosteal incisions were performed to allow for passive closure of the graft prior to

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**Fig. 7** – The block is carefully shaped to assist in stable and close adaptation to the recipient bed. Care should be taken to avoid over-reducing the bone block graft. If excessive changes are needed to achieve stability, then the recipient site should be adjusted as well.

**Fig. 8** – The block is fixed with at least two fixation screws to ensure stability and anti-rotation. Screw osteotomies through the block should be lagged so that threads do not engage. Intra-marrow penetration is performed under the block, at the recipient bone bed, prior to fixation. The gap was filled with particulate bone graft (left maxillary site).

**Fig. 9** – The same procedures were carried out in the right maxillary site.

**Fig. 10** – Two bone blocks were fixed to the underlying bone forming a room with vertical and horizontal walls.
placement of additional particulate graft material. Typically conventional periosteal releasing incisions are adequate for closure; the surrounding area of the block is then mortised with a particulate bone graft.1

Closure of the recipient site is critical to success in block grafting. Wound dehiscence at the recipient site has been associated with more block resorption or complete loss of the grafted bone.

All loose teeth and rootless teeth were extracted before closure.

The everted wound margin and the ridge crest is then secured with interrupted sutures along its length at regular intervals.

Postoperative medications are aimed at control of pain, swelling, and infection is recommended for up to 5 days post-operatively. Antibiotics in the form of Amoxicillin-Calavulonic acid were started immediately before the surgery and continued for 5 days twice daily. Also, 100 mg of hydrocortisone was administrated immediately preoperative, and a long acting steroid was administrated after recovery. NSAD was prescribed for pain control. Also, chlorhexidine mouth wash was started from the second day after surgery.

Sutures were retained in place as long as they appear to be providing wound stability, which in this case were kept 14 days.

7. Implant placement

After successful initial healing, ramus block grafts was allowed to mature for 4 months prior to uncovering and implant placement [8,16,17]. During the healing phase the block integrity was evaluated radiographically (Figs. 11 and 12).

The patient was admitted to a hospital and general anesthesia was administered. Reentry to the block site required re-incision of the overlying tissue, following the same incision lines used in the first surgery. The block stability was assessed clinically and fixation screws removed. The bone grafts were taken by the recipient sites; however some resorption in the graft especially in the maxillary right and left lateral incisors was noticed.

Knife-edge ridges and any bone irregularities were flatted to allow a thickness of at least 4 mm. Screw-shaped implants with sandblasted acid-etched surface, 3.75 to 4.1 mm diameter and 8–11 mm length, were used in this study. The preparation of implant sites was carried out with twist drills of increasing diameter under constant irrigation. Implants were positioned at the bone crest level. Care was taken to assess the position of the mental foramen. Implant sites were marked using a surgical template. The templates were based on the diagnostic wax-up with perforations on the longitudinal axis, on the premolar and molar regions, according to ideal position of final implant-supported restorations (Figs. 13 and 14).

With the aid of the pre-operative surgical implant guide, it was possible to insert the implants in their anticipated position. Ten tapered design implants2 were inserted. All have S.L.A surface that provide higher bone-to-implant contact and faster bone formation on the surface.

Four implants were placed in maxillary right side. The implant placed in the lateral incisor was Narrow Ridge type.3 Two implants were placed in the left side. One Narrow Ridge (NR) type (3.00 mm) in lateral incisor and one regular (3.75 mm) in first molar area were inserted. Tow implants were placed in the right mandibular canine and first molar area. Also, two implants were placed in the left mandibular canine and first molar area.

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1 Bio-oss (Geistlich Pharmaceutical, Wolhusen, Switzerland).
2 Superline, Dentium Implant Co. F209, 107, Gwanggyo-ro, Yeongtong-gu, Suwon-si, Gyeonggi-do, Korea 443-270.
3 NR Line Body Ø3.1, Platform Ø4.3.
Primary wound closure was achieved with horizontal mattress sutures alternated with interrupted sutures to ensure a submerged healing procedure in dental implants. After 6-month healing period, the implants were exposed under local anesthesia and abutments were connected to the implants. The abutments were prepared under constant irrigation to insure that the abutments were parallel to common path of insertion.

The remaining natural teeth were also, prepared to receive porcelain-fused to metal crowns on the maxillary incisors and six Units Bridge on the two mandibular canines.

A direct impression technique was used in which impression is made to the prepared abutments in the patient’s mouth. A dowel impression technique with silicone was made after blocking the insertion holes on top of the abutments. Jaw relation records were obtained to mount the casts. The obtained impressions were poured in two stages. The first included pouring the abutment portion with acrylic resin. A wire was inserted in the acrylic resin before setting to make mechanical interlock with the second stage which was poured in improved stone. This was made to insure preservation of the cast from scratching during the different procedures of prosthesis manufacturing.

On the poured casts, with the assistance of a scanner, three-dimensional data are produced on the basis of the master die. These data are processed by means of dental design software. After the CAD-process the data were sent to a special milling device that produces the real geometry in the dental laboratory. Finally the exact fit of the framework was evaluated and, if necessary, corrected on the basis of the master cast. The ceramist carries out the veneering of the frameworks in a powder layering or over pressing technique\(^5\)\(^6\) (Figs. 15–17).

After try-in of the milled substructure in the patient’s mouth, the resin was invested and cast to metal. Porcelain build-up was carried out and baked. A porcelain try-in in the patient’s mouth was made and margins, embrasures and occlusion were checked. Esthetics and phonetics required few modifications. The final prosthesis composed of many sections.

In the maxilla, the two centrals were separate. A two unit's bridge restored maxillary right first and second premolars. Another two unit's bridge restored the right first and second molars. In the left side, Three-units Bridge restored the first, second premolar and first molar.

In the mandible, one six unit's bridge restored the space from canine to canine. Two three units Bridge restored the first, second premolar and first molar.

The final prosthesis restored the esthetic of the patient and provided lip support. It also, provided fullness of the check. The contour of the maxillary anterior teeth was made to follow the smile line of the lower lip and enhanced natural look. With a slight horizontal overlap, it was possible to mask the moderate class III that was present in the start of the treatment.

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\(^4\) Duralay, Reliance Dental Mfg. Co, USA.

\(^5\) CAD/CAM SHERA Werkstoff-Technologie GmbH & Co. KG, Germany.

\(^6\) Dental Wings software, 2251 Letourneux, Montreal, QC H1V 2N9, Canada.
The patient pronunciation of many sounds improved immediately after insertion of the prosthesis. The patient was able for the first time in her life to chew certain types of foods that she never chewed before (meat and salads) (Figs. 21 and 22). This raised the patient’s self-esteem and confidence among her dental colleagues, friends and family.

8. Discussion

Dental implants have become an accepted treatment modality for aging patients with either completely or partially edentulous arches [19]. However, in partially edentulous children who have ED, multiple implant placements are not possible because of the ongoing development of the jaws and insufficient bone. In addition, the bone height and width will not be sufficient for implant insertion without advanced surgical approaches [20]. In patients not treated with ED, craniofacial deviations from the norm increased with increasing age with a tendency towards a Class III pattern with anterior growth rotation [21].

In this patient, she does not have any of the manifestations of ED except that Partial Anodontia and severe atrophy of alveolar ridges and protuberant lips. All other tissues of ectodermal origin were normal. She have got good long hair, well lined eye brow, long eye laches, healthy nails and normal skin [4].
There are several reports of successful dental implant reconstruction in patients with ED. Although most of reports have addressed implant reconstruction in the severely atrophic maxilla, few have addressed reconstruction of the both maxilla and atrophic mandible [2,22]. Further, only a few authors report the use of implants in the treatment of adult patients with ED and reports on bone augmentation are lacking [2,23]. There is also, a paucity of published articles on 3Ds building-up a room of two walls (vertical and horizontal) and fill it with particulate bone to augments severely resorbed ridges in ED patients.

Ridge augmentation procedures prior to conventional fixed prosthodontics or implant therapy are indicated when an adequate width or height of the alveolar ridge is not present. Overall the survival rates of implants placed in augmented ridges is 87% (range from 60% to 100%) [11]. The present case had severe bone atrophy in the maxilla and mandible due to congenital ED. Thus augmentation was necessary to place the implants in a biologically accepted and prosthetically driven location to achieve optimal function and esthetics [24].

The use of corticocancellous bone grafts for ridge augmentation in implant dentistry was first reported by Breine and Branemark [25]. The revascularization of cortico-cancellous block grafts takes place at a much faster rate than in cortical bone autografts. Revascularization of block grafts enables maintenance of their vitality, and, hence, reduces chances of graft infection and necrosis [6].

Autograft is considered as the Gold Standard for bone transplantation [6] and various studies have shown efficacy for same [25–35]. It is osteogenic, osteoconductive and osteoinductive. Autografts can be derived from extra oral source (iliac crest, ribs) or intraoral source (chin, ramus). They can be used in block or particulate form [6]. Corticocancellous block grafts are preferred because of enhanced revascularization of the cancellous portion, and mechanical support and

Fig. 21 – Panoramic radiograph showing the full mouth implants rehabilitation in the grafted ridges.

Fig. 22 – CBCT showing the implant supported prosthesis within the grafted block.

Fig. 23 – Side view of the patient after insertion of the prosthesis. It provided lip and cheek support.
rigidity of the cortical portion, which ensures optimal ridge augmentation [31]. The healing of autogenous block grafts has been described as “creeping substitution” where viable bone replaces the necrotic bone within the graft and is highly dependent on graft angiogenesis and revascularization [6]. There is no risk of rejection or adverse immunological reaction with autogenous bone grafts. They are highly advantageous but are associated with some risks, such as donor site morbidity, limited bone availability, size mismatch, drooping of chin, nerve damage, tooth devitalization, gingival recession, increased postoperative discomfort, infection and blood loss [32].

In this case, autogenous corticocancellous block graft from ramus region was obtained of same surgical site, thus avoiding secondary site morbidity and used it for vertical and horizontal ridge augmentation in right side. The same was done in the left side. Bleeding points were created on recipient bed, which increases rate of revascularization, the availability of osteoprogenitor cells and the rate of remodeling. Block graft was stabilized using titanium screws to avoid movement. The key to success is elimination of graft mobility and dead space between the graft and host bone [32,33].

Block grafts are associated with minimal resorption and do not usually require the use of an overlying membrane unless the dimensions of the graft are inadequate. Block grafts take longer to integrate than cancellous bone grafts. When a block graft is used, a staged surgical approach is recommended as opposed to placing the implants in conjunction with the graft [34]. The mandibular ramus is a useful, cortical graft that provides primarily dense. In addition, the mandibular ramus donor site is associated with fewer postoperative complications, in comparison to the symphysis region. Hence they can be successfully used for alveolar ridge augmentation prior to implant placement [35,36].

The mandibular second molar area provided the thickest cortical graft averaging range 2.0–4.2 mm. A cortical plate of 2.8 mm in average is enough to augment large defects [37]. This ramus block in one side provided enough bone to build horizontal and vertical walls in both right quadrants of the maxilla and mandible. The other side was enough to build the left side quadrants.

The present study showed that intraoral bone block graft surgery is a predictable operation for the use of dental implants. This procedure offered additional bone for dental implant placement. The high implant survival and radiologic success shown can be attributed, among other factors, to the improvement of the crown-implant ratio. The higher amount of available bone following augmentation allows placement of longer implants in a better trajectory.

In a longitudinal study [36] in which the Follow-up from dental implant placement ranged from 6 to 67 months (mean: 24.3±11.2 months). The overall survival rate was 96.9%. The 5-year cumulative survival rate was 88%, Marginal bone loss around implants ranged from 0 to 3.3 mm (average: 0.22–0.45). Only 5% of the implants presented marginal bone loss more than 1.5 mm during the follow-up time [37–42].

The prosthetically-driven implant placement of the implants allowed placement of the implants in a position that allowed restoring the patient esthetics and functions and raised her self-esteem.

CAD/CAM technologies have started a new age in dentistry. The quality of dental prostheses has improved significantly by means of standardized production processes. The CAD/CAM milled substructure allowed accurate making and checking of the final prosthesis. It also, allowed modifications of the substructure by the soft wear and eliminated the need for remake of the fabrication procedures in case modifications, addition or alterations were required [7,43].

9. Conclusion

This article presents a case of full mouth implant rehabilitation in severe alveolar ridge atrophy associated with congenital ED in young female. Augmentation in a partially edentulous patient prior to implant placement, using autogenous bone grafts harvested from the mandibular ramus and firmly secured to the recipient site with osteosynthesis screws was carried out. The detailed procedures of surgical and prosthetic management were described with special emphasis on the precautions and risk factors.

The clinical indication for the case described was the lack of sufficient alveolar bone quantity, a situation that could interfere with the esthetics and functional loading of implants. The mandibular ramus block bone grafts gives predictable outcome within a short healing time and provides ideal sites for endosseous implant.

Applying the principles of prosthetically-driven implant placement, it was possible to provide the patient with an
implant supported prosthesis that is successfully restored the patient lost esthetics and functions.

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