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Case Report

Dentistry Section

Ectodermal Dysplasia: Management of Knife-Edged Irregular Ridge and Its Rehabilitation with Hybrid Implant Prosthesis

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ABSTRACT

Ectodermal Dysplasia Syndrome (EDS) is mainly X-linked inherited disorder with male predominance. According to Lyon hypothesis, female patients may show partial expression of EDS. Oral findings include hypodontia, rarely anodontia, protuberant lips, hyposalivation, conical teeth and loss of vertical dimension. The alveolar process fails to develop in the three dimensions. Such patients present a challenge to dental treatment due to an irregular residual ridge. This case report presents oral, functional and aesthetic rehabilitation of a 21-year-old female diagnosed with EDS using implants in the anterior maxilla at the sites of the canines bilaterally. Following, implant placement, it was noted that implant in the region of right canine was labially inclined compared to implant in the region of left canine, but both were centered in the ridge. To manage non-parallelism, one-piece titanium framework was fabricated using computer numeric controlled (CNC) machine. Subsequently, tooth and gingival shade ceramics were fired to simulate natural teeth and compensate for gingival deficiency respectively. The treatment described here restored patient's aesthetics, function, self-confidence and status in society.

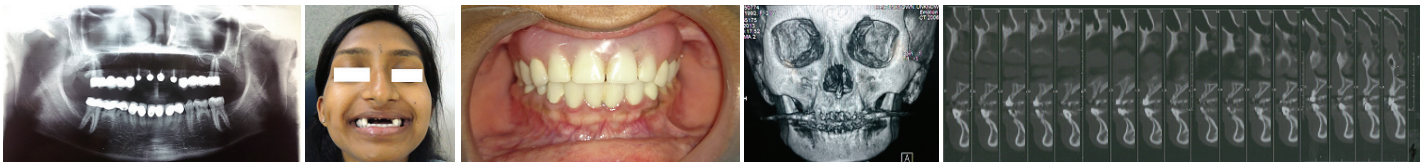
Keywords: Aesthetics, CAD-CAM, Ectodermal dysplasia syndrome, One-piece titanium framework

CASE REPORT

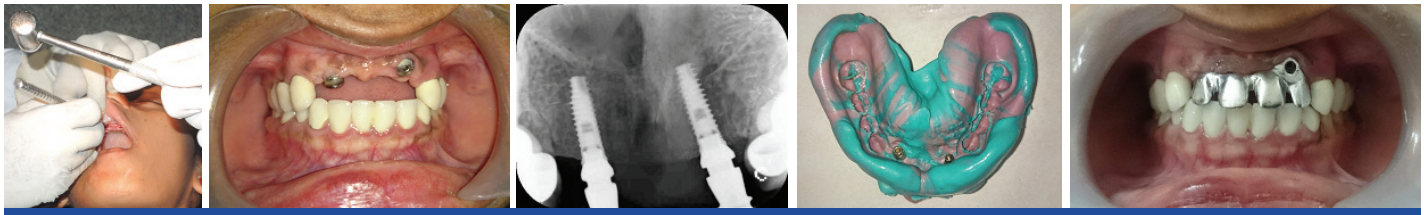
A 21-year-old female presented to the Department of Prosthodontics with the chief complaint of an unattractive smile. On oral examination, teeth present were 16, 14, 53, 51, 61, 63, 24, 25 in the maxillary arch and 36, 75, 34, 33, 32, 31, 41, 42, 43, 84, 85, 46 in the mandibular arch. Orthopantomogram revealed root canal treated 53, 51, 61, 63 with short root length and bone loss around the cervical third of roots [Table/Fig-1]. The treatment plan was formulated to extract 4 deciduous maxillary anteriors [Table/Fig-2] and insert an interim acrylic resin labial flange removable partial denture to restore aesthetics [Table/Fig-3]. CT scan of the patient revealed class IV maxillary ridge according to Cawood & Howell with a bone thickness of only 2 mm in the cervical region. Labial and palatal cortical plates were intact [Table/Fig-4,5]. Definitive treatment included alveolar bone splitting and immediate implant placement in the canine region bilaterally after 1 month of extraction.

Full thickness mucoperiosteal flaps were raised on the labial and palatal aspects of cortical plates with minimal tissue reflection on palatal aspect to preserve the periosteum attachment to surrounding bone, maintaining blood supply to the area of implant placement and facilitating healing. Knife-edged ridge was smoothed with a large round bur to eliminate any sharp bony spicules and provide a base for the osteotome guided alveolar split osteotomy. Horizontal osteotomy cut was made to a depth of 3 – 4 mm using a thin fissurotomy bur; 2 mm away from the first premolar on adjacent sides facilitating osteotome guided alveolar split osteotomy. The sharp tip of osteotome was inserted through the horizontal cut and gentle tap with a surgical mallet was given to gradually expand the ridge [Table/Fig-6]. Implant sites were then prepared in the canine region bilaterally, with final twist drill of diameter 2.0 mm keeping drill hole centered labiopalatally and following ridge morphology. Implants of size 3.3 x 13 mm (Classix, Premium, Cortex Dental Implants Industries Ltd, Shlomi, Israel) were inserted. Healing collars were secured in place and alloplastic bone graft material (Perioglas, Novabone, Jacksonville) was used to fill any bone defect present. The mucoperiosteal flap was repositioned back and tension-free suture closure was performed using 3-0 silk suture. The acrylic resin labial flange removable partial denture was checked and modified to avoid any contact with the healing collar of implants and inserted. After a healing period of 1 week, sutures were removed.

Recall appointment was given after 6 months. It was noted that implant in the region of 23 was labially inclined compared to implant in the region of 13 but both were centered in the ridge [Table/Fig-7]. IOPA radiograph revealed osseo-integration of implants with bone [Table/Fig-8]. Healing collars were unscrewed and transfer impression copings (Classix, Premium, Cortex Dental Implants Industries Ltd, Shlomi, Israel) were attached to both the implants. A verification stent was fabricated by adapting low shrinkage pattern resin (Pattern Resin LS, GC, America) around mid-section of transfer copings, connecting them to minimize distortion and record an accurate impression. It should be thick enough to minimize flexure during insertion and removal. The stent was removed and re-inserted to verify passive fit. Light body addition silicone impression material (Aquasil ultra, Dentsply, York, PA, USA) was injected under the stent and around soft tissue surrounding implants while the custom tray was loaded with vinyl polysiloxane soft putty impression material (3M ESPE, USA) and light body addition silicone impression material (Aquasil Ultra,



[Table/Fig-1]: Pre-treatment orthopentomogram depicting oligodontia and presence of multiple deciduous teeth. **[Table/Fig-2]:** Frontal extraoral view demonstrates compromised aesthetics. **[Table/Fig-3]:** Intraoral view of interim acrylic resin labial flange removable partial denture inserted to restore aesthetics until definitive treatment with implants. **[Table/Fig-4]:** Computed Tomography (CT) scan demonstrates irregular bony contour in premaxilla region. **[Table/Fig-5]:** Available bone width of 2mm in cervical region of the premaxilla on a CT scan.



[Table/Fig-6]: After initial horizontal osteotomy cut with fissurotomy bur, sharp tip of osteotome was inserted through the cut and gentle tap was given by a surgical mallet to expand the alveolar ridge. **[Table/Fig-7]:** Intraoral view demonstrates labially inclined implant in the region of left canine compared to the implant in the right canine region due to irregular bony contour. Both implants were centered labiolingually in the ridge. **[Table/Fig-8]:** IOPA radiograph reveals osseointegration of implants with bone, 6 months after placement. **[Table/Fig-9]:** An open custom tray impression was recorded after verification stent was fabricated connecting both transfer impression copings. **[Table/Fig-10]:** One-piece titanium framework trial insertion to verify fit intraorally.

Dentsply, York, PA, USA). A custom open tray impression was then recorded. Transfer impression copings were unscrewed and the impression was removed after recommended period of setting time according to manufacturer's guidelines [Table/Fig-9]. Lab analogs were attached to both impression copings and the impression was poured with Type 4 dental stone (Kalabhai, Mumbai). The cast was removed from impression after recommended period of initial setting time. Transfer impression copings were unscrewed from the cast. A resin pattern (Pattern Resin LS, GC, America) was fabricated to simulate the design of final one-piece titanium framework. It was laser scanned and the image was transferred to computer-aided design software on a computer. After required design modifications, the framework was milled from a block of grade-2 titanium in a computer numeric control (CNC) machine (Amann Girrbach America, Inc. Charlotte, NC, USA). The milled framework was then finished and polished by a technician. The one-piece titanium framework was tried-in the patient to verify passive fit over the implants by using alternate finger pressure and absence of screw resistance [Table/Fig-10]. There were no interferences noted from the opposite dentition. The framework was sandblasted and acid etched to receive ceramic veneering. Ceramic layers (Creation CC, Jensen Dental, North Haven, Connecticut) were fired and final glazing and polishing was done to simulate natural tooth structure. Gingival ceramic (Creation CC, Jensen Dental, North Haven, Connecticut) was fired and glazed to simulate marginal and attached gingiva. The exposed surfaces of titanium framework were finally polished and the prosthesis was inserted and torqued to a preload of 35 Ncm onto the implants. The access holes were sealed with glass ionomer cement [Table/Fig-11]. The patient was satisfied with aesthetics and follow up appointment after 6 months revealed healthy peri-implant tissue without any implant superstructure complications [Table/Fig-12].

DISCUSSION

Ectodermal dysplasia syndrome (EDS) is a large, heterogeneous group of inherited disorders in which two or more ectodermal derived anatomic structures fail to develop. Hypoplasia or aplasia of ectodermal appendages occurs, which includes skin, hair, nails, eccrine glands, and teeth [1-3]. Since, it shows an X-linked inheritance pattern, male predominance is seen, though female patients may show partial expression of EDS that can be explained by Lyon hypothesis.

[Table/Fig-11]: Tooth and gingival shade ceramic were fired to produce definitive prosthesis and prosthesis secured in place Intraorally. **[Table/Fig-12]:** Extraoral view of completed case shows restored function and aesthetics.

Oral manifestations are defined by hypodontia (missing five or fewer teeth), oligodontia (missing six or more teeth), and rarely anodontia (missing all teeth); truncated or cone-shaped teeth, hyposalivation due to salivary gland involvement. Lips may be dry and cracked with pseudorhagades formation. High palatal vault and cleft palate may be present. The alveolar process fails to develop in sagittal, coronal and vertical dimensions resulting in a knife-edged ridge. However, even with anodontia, the growth of the jaw is not impaired [4-6].

Oral rehabilitation in the skeletally mature adult can be in the form of conventional fixed and removable partial dentures or more modern implant therapy [6,7]. The presence of conical teeth can complicate retention for fixed partial denture while reduced knife-edged ridges present problems in the form of support and retention for a removable prosthesis [8]. There is a risk of further bone loss in a comparatively young individual due to conventional treatment. Implant therapy can help overcome these shortcomings and restore function, aesthetics, and phonation.

However, in patients with EDS, deficient width of alveolar bone poses a problem. Several techniques have been proposed for implant placement in patients with severe atrophy of the maxilla in the horizontal dimension [9]. Osborn described the 'extension plasty', which is a two- stage technique for splitting and extending the alveolar crest. The expanded space was filled with hydroxyapatite or autogenous bone. Second stage surgery was performed 8–12 weeks later to insert the implant [10]. Nentwig & Kniha, in 1986, described a bone splitting technique that allowed expansion of the alveolar crest and placement of the implant at same time unlike 'extension plasty' described by Osborn [11]. Summers RB described a technique called Ridge expansion osteotomy (REO) which maintains the available bone by pushing the bone adjacent to the osteotomy site with the osteotomes, resulting in bone compaction. This technique is, however, ruled out in cases where the labiopalatal thickness of bone is less than 3 mm [12].

Alveolar bone, deficient in all three dimensions may prevent placement of implants parallel to each other, which is otherwise desirable. In such cases, CAD/CAM fabricated one-piece titanium framework present the advantages of being specific to the patient and a passive fit that otherwise, could not have been achieved. Traditionally, lost wax technique and casting has been used to fabricate implant superstructures. However, the technique employed is cumbersome and leads to casting errors. The framework may not fit accurately or passively on the implants. In addition, casting defects in the form of micro-porosities weaken framework leading to failure under loading. CNC milled frameworks are accurate with significantly less distortion [13-15].

Acrylic resin and gingival shade composites are still in use to replicate natural teeth and gingival tissues respectively. However, wear is comparatively faster with these materials. In addition, porous structure of resin leads to discoloration and absorption of oral fluids [13]. Tooth shade and gingival shade ceramics provide a better solution to the above-mentioned problem.

CONCLUSION

The technique described addresses problems associated with the bone contour which results in non-parallel implants and discusses best treatment option available at present. The treatment described here restored patient's aesthetics, function, self-confidence and status in society.

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