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Case Report Article

Prosthetic rehabilitation in a patient with Singleton Merten syndrome and acrylic resin hypersensitivity

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Abstract

Introduction: Singleton Merten Syndrome is a rare disease characterized by the presence of the dental dysplasia phenotype, calcifications in the aorta, progressive wear and loss of bone protein (osteoporosis) in the hands and feet. Patients have muscle weakness, poor motor development, abnormal dentition, deformities of the feet and hands, and skin lesions. **Objective:** This report describes the maxillomandibular rehabilitation of a patient with Singleton Merten Syndrome and an allergic reaction to the acrylic resin through maxillary overdenture and mandibular partial removable denture. **Case report:** Female patient, 18 years old, with clinical characteristics of Singleton Merten Syndrome and allergic reaction to acrylic resin, with complaints of loss of function and esthetics due to the absence of several teeth, but with the presence of unerupted maxillary and mandibular tooth buds. Maxillary overdenture and mandibular removable partial denture were made of polyethylene. **Conclusion:** The rehabilitation treatment with maxillary overdenture and mandibular partial removable denture provided better facial muscle support, restoring masticatory function and facial esthetics. With this treatment, it was possible to reestablish patient's satisfaction and self-esteem due to the correct construction of the prostheses, and minimize hypersensitivity reactions in the oral mucosa, which allowed the use of these prostheses by the patient without any complications.

Introduction

Singleton Merten syndrome (SMS) is an autosomal dominant genetic disorder with variable expression and symptoms present during childhood [4, 5, 13, 17]; the pathophysiology is unknown because of the rarity of this disease [5, 12, 16]. SMS major features are extensive aortic calcifications, dental anomalies due to the phenotype of dental dysplasia, abnormal ossifications, psoriasis, and glaucoma [12].

Clinically, individuals with SMS show with fever of unknown origin, muscle weakness, impaired motor development, abnormal dentition, glaucoma, photosensitivity, heart block, deformities of the feet and hands, skin lesions, and chronic psoriasis [2, 5, 16]. Radiological aspects include skeletal demineralization, expanded shafts of metacarpal and phalanges with extended medullary sinuses, cardiomegaly, and proximal aortic intramural calcification, with occasional extension to the mitral or aortic valve [2, 16, 19].

Based on dental development, the main features are developmental anomalies observed as late exfoliation of molars; late eruption of premolars; formation of truncated or missing roots in incisors, canines, and molars; and abnormal alveolus with extensive area of bone resorption [5].

Thus, prosthetic rehabilitations as overdentures, removable partial dentures (RPD) and implant-supported prosthesis are indicated for those cases [20]. The overdenture are removable dentures that cover the retained teeth, roots dental, or implants allowing an additional retention to the alveolar edge, recovering the function and esthetics [1, 10]. The most widely used material for overdentures is acrylic resin, with physical, mechanical, and esthetic properties. However, acrylic resins can cause hypersensitivity reactions with pain sensation and oral burning in some patients, resulting in allergic stomatitis over the prosthesis [14, 20]. Because of these reactions by the use of acrylic resin-based prostheses, researchers seek more biocompatible materials, such as polyethylene, light-cured urethane dimethacrylate (UDMA) composite, for covering the prosthesis, in addition to laboratory maneuvers as microwave curing cycle, warmed aqueous environment, or higher temperatures under pressure [20].

The literature is scarce in relation to the rehabilitation treatment with overdenture on roots in patients with Singleton Merten syndrome. Additional studies are needed to elucidate this kind of treatment in these patients. This, this case report shows the rehabilitation treatment of patient with clinical features of Singleton Merten syndrome and allergic reaction to acrylic resin through maxillary overdenture and mandibular RDP.

Case report

Patient T. M. E, female, 18 years-old, was referred to the Clinic of Dentistry of the University of Ribeirão Preto searching rehabilitative treatment to return esthetic due to some missing teeth and inadequate formation of other.

At anamnesis, the patient revealed she had hereditary rheumatoid arthritis in the hands and feet, cutaneous pigmentations (figure 1 – A and B), syndrome of Singleton Merten, and allergic reaction to acrylic resin-base prosthesis, evidenced previously.

The panoramic radiography showed included (#51, #52, #11, #13, #14, #15, #18, #61, #62, #21, #22, #23, #24, #25, #27, #28) and erupted maxillary teeth (#16, #17, #26, #53, #54, #55, #64, #65). In the mandible, the radiograph revealed the tooth agenesis (#31, #32, #41, #42), retained teeth (#73, #75, #83, #85, #34, #38, #44, #47, #48) and erupted teeth (#36, #37, #46) (figure 2). The established clinical diagnosis was of partial maxillary/mandibular edentulism associated with reduced vertical dimension, in function of the incomplete eruption of maxillary/mandibular posterior teeth, and proper maxillomandibular relationship (figure 3 – A and B).

Figure 1 – Hereditary rheumatoid arthritis and cutaneous pigmentations on: A) hands and B) feet

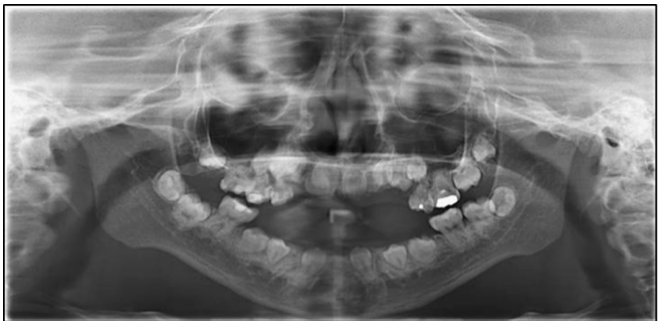

Figure 2 – Panoramic radiograph

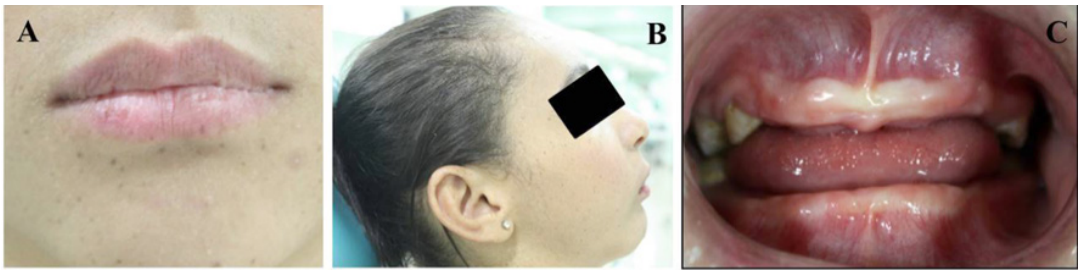

Figure 3 – A) Initial Extraoral clinical aspect (frontal view); B) Initial Extraoral clinical aspect (profile); C) Initial intraoral clinical aspect

To establish the proper treatment plan, we performed the impressions of the maxilla and mandible with the aid of alginate (Jeltrate Plus - Dentsply, Petrópolis, RIO DE JANEIRO, Brazil), for obtaining the study models (figure 4 - A and B), in type III dental plaster for the maxillary dental cast and type IV for dental plaster for the mandibular dental cast (Herodent and Herostone - Vigodent, Rio De Janeiro, RIO DE JANEIRO, Brazil). The dental casts were assembled a in semi adjustable articulator (SAA) (BioArt, Sao Carlos, SP, Brazil) at centric relation, with the aid of acetate articular plates and wax roller for the correct determination of the orientation plans, to evaluate the intermaxillary relation and the available prosthetic space.

After planning, the proposed treatment was maxillary overdenture because of the presence of short and expulsive crowns of the posterior teeth that would prevent the adequate RPD retention. In mandible, RPD was planned. Both prostheses were constructed in polyethylene because of the patient's allergic reaction to acrylic resin. In the light of oral and systemic limitations presented by the patient only this treatment option was proposed. The plan was accepted by the patient.

A customized tray was constructed in acetate (acetate plate, BioArt, São Carlos, SP, Brazil), extending throughout the prosthetic area and 2 mm below the bottom of the vestibule. Next, the working impression was performed through the peripheral sealing with green Godiva stick (Kerr, Joinville, SC, Brazil). The functional impression was performed with Soft Impregum (3M, Sumaré, São Paulo, SP, Brazil).

To obtain the mandibular working cast, the study cast was delineated (Delineador B2-Paralelômetro, BioArt, São Carlos, SP, Brazil), to allow the planning for the determination of the characteristics of the retention clasps and reciprocity and location of niches for the RPD construction. After the planning and design, the niches on the mesial side of the occlusal face of teeth #46 and #36 were performed and the teeth molding with alginate (Hydrogum 5, Zhermack SpA, Badia Polesine, RO, Italy).

After the obtaining of the maxillary and mandibular working casts, an acetate proof base and wax roller was made (Figure 5-A and B) for the construction of guidance plans. Through the maxillary proof base, we determined the height of the upper guidance plan, lip support and buccal corridor and, later, the parallelism between occlusal plane and bipupilar line and between the occlusal plane and Câmper plane (Figure 6-A and B). Then, with the aid of facial arch (BioArt, São Carlos, SP, Brazil) the maxillary working cast was mounted on SAA.

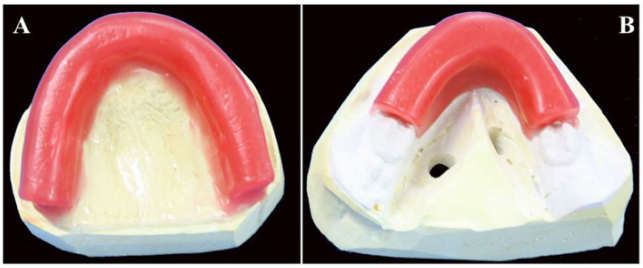

Figure 5 – A) Acetate-based proof base and wax roller (maxillary working cast); B) Acetate-based proof base and wax roller (mandibular working cast)

Figure 6 – Guidance of the maxillary wax plane A) front view: parallelism between occlusal plane and bipupilar line; B) lateral view: parallelism between the occlusal plane and Câmper plane

Following, the vertical dimension was determined by the association of the metric, phonetic and aesthetic techniques through Dawson's central relation bimanual manipulation, and the reference points (high smile line, canine-to-canine distance, and midline) were marked to allow the election of artificial teeth (figure 7 - A, B, and C).

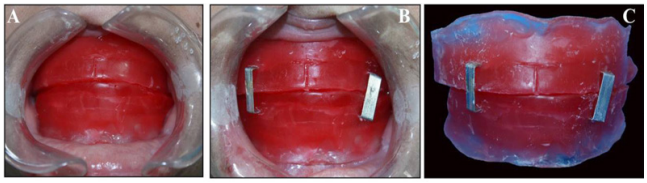

Figure 7 – A) Determination of the vertical dimension of occlusion; B and C) register in central relation and reference points for election of artificial teeth

The mandibular working cast was mounted on SAA, and the models and planes of orientation sent to the prosthetic laboratory for the assembly of artificial teeth. The tooth shade previously selected was 61 of the Biotone color scale (Dentsply, Petrópolis, RIO DE JANEIRO, Brazil) (figure 8).

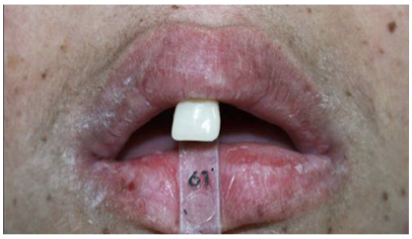

Figure 8 – Color selection of artificial teeth

After the assembly of teeth in wax (figure 9), the esthetic and functional proof was carried out (figure 10 – A and B).

Figure 9 – Fitting of the teeth on SAA

Figure 10 - A) Assembly of teeth in wax; B) esthetic and functional proof of teeth on wax, intraoral aspect

After obtaining the prosthesis through the exothermic polymerization reaction of the material (polyethylene) in the laboratory (figure 11), the prosthesis received finishing and polishing, and then were installed and adjusted, restoring 's function, phonetics, and aesthetics (figure 12).

Figure 11 – Internal view of the finished prosthesis. A) Maxillary overdenture ;) B) Mandibular RPD

Figure 12 – Installed prostheses

Discussion

According to the literature, the use of osseointegrated dental implant is considered the rehabilitation treatment of choice for patients with Singleton Merten syndrome [8, 16]. According to Rodriguez et al. [16], the result of the treatment depends on the local bone density and volume,

often requiring bone grafts, but there is a positive correlation between the success of rehabilitation treatment and dental implants [7].

In this study, the installation of osseointegrated implants were not possible because of the presence of deciduous and permanent tooth buds, which can be considered as a contraindication; and the patient's age because the growth of the maxilla and mandible was still active. Thus, the rehabilitation treatment of choice was the installation of a maxillary overdenture and mandibular RPD.

The literature reports that overdentures are an advantageous option for edentulous patients with short tooth crown and without retention, absence of eruption of teeth, and difficulties in adjusting to the prosthesis, making possible to restore the esthetics and physiological functions of the oral cavity. Compared to conventional dentures, overdentures feature advantages: better support, stability and retention, ease of phonation, more comfort during the function, and maintenance of volume and height of alveolar bone [3, 10, 11, 18].

For the fabrication of total and removable dentures, the material of choice most commonly used is the acrylic resin, but this material may cause hypersensitivity and allergy due to the occupational contact of composites as HEMA, EGDMA and TEG-DMA with oral mucosa [14, 20]. Allergic stomatitis under the prosthesis, for example, is related to the use of mucous-supported dentures, thanks to the presence of the sensitizing substance, methyl methacrylate [6]. According to Kedjarune et al. [9], greater amounts of methyl-methacrylate are incorporated in the saliva of the wearers of prosthesis confectioned in acrylic resin, causing sensation of local pain and heat. To minimize hypersensitivity reactions overdentures made of nylon, silica, and polyethylene are indicated [15, 20].

The polyethylene is a partially crystalline, flexible polymer, used as a material of structural reinforcement in the dentistry practice for confection of total and removable prostheses. It presents advantages as single body formation, polishing easiness, satisfactory esthetic, flexural resistance, possibility of reassembly, adhesiveness, and minimizing reactions of hypersensitivity in oral mucosa [20, 21]. The main disadvantages are possible dimensional changes during polymerization and porosity [22].

In this present case report, the patient was allergic to acrylic and then we choose polyethylene to construct the prostheses. However, the literature lacks on the relation of hypersensitivity to acrylic and the Singleton Merten syndrome.

Conclusion

The treatment through maxillary overdenture and mandibular removable partial denture rehabilitated facial muscle support, masticatory function, and esthetics of teeth and smile. With this treatment, it was possible to return the patient's satisfaction and self-esteem.

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