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A RARE CASE OF ASYNDROMIC SEVERE HYPODONTIA IN 3 SISTERS

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ABSTRACT
Tooth agenesis is the most common dental anomaly and the occurence of these anomalies in more than one family member suggests a definite role of genes in hypodontia. This article describes a rare case of severe non-syndromic hypodontia in three sisters and highlights the associated oral and paraoral findings. Hypodontia can have profound psychosocial impact in the daily lives of the patients. Hence early diagnosis and intervention is very important. After discussion of various treatment options available and considering the financial status, conventional overdentures were constructed for the eldest sister which greatly improved her function and appearance.

KEYWORDS: Hypodontia, treatment options, prosthesis.

INTRODUCTION
The term 'hypodontia' refers to the developmental absence of one or more teeth, either in primary or permanent dentition, excluding the third molars.[1] It is the most common dental anomaly with the prevalence of 2.6% to 11.3%.[2] Accurate diagnosis of hypodontia depends on radiographic, clinical and dental cast examination to determine whether the tooth is extracted, impacted or congenitally absent.[3]

Hypodontia is found to be more common among persons who are genetically related.[4] Females are affected more frequently than males by a ratio of 3:2.[5] Absence of one or two teeth is relatively common whereas anodontia, which is the developmental absence of all teeth is a rare finding. Absence of six or more teeth is termed as severe hypodontia and it maybe associated with a syndrome like ectodermal dysplasia.[6] Hypodontia may be linked with various dental anomalies like microdontia, delayed eruption and exfoliation of teeth, taurodontism, hypocalcification, lack of alveolar development and class 3 skeletal relationship.[7] Hypodontia has significant clinical implication as it can seriously affect a person's emotional and physical well being.[8]

Hypodontia is considered as a multifactorial condition with both genetic and environmental influences. Muscle specific homeobox genes namely, Msx1 and Msx2 involved in the tooth development are reported to be mainly affected.[2] Environmental factors include trauma in the dental region, surgical procedures, extraction of preceding primary tooth, infection during pregnancy and early childhood, drugs like thalidomide, hormonal and metabolic influences, radiation therapy, chemotherapeutic agents and evolutionary trend towards fewer teeth.[8]

Various treatment options available for hypodontia patients include orthodontic movement and/ or restorative replacement in form of dentures, crowns, bridges, autotransplantation and dental implants.[9] The choice of a particular treatment option depends on factors like age of the patient, number of missing teeth, adjacent teeth condition and position, amount of bone resorption and medical factors and financial status of patient.[10]

CASE REPORT
A set of 3 sisters presented to the Department of Prosthodontics, de'Montmorency College of Dentistry/Punjab Dental Hospital, Lahore with the chief complaints of multiple missing teeth in the eldest sister and prolonged retention of primary dentition in the 2 younger sisters. Patients are identified as case 1, 2 and 3.

Case 1
The eldest patient was 18 years old female, resident of Kabul, Afghanistan with presenting complaint of missing teeth which caused difficulty in chewing and gave an aged appearance to the patient. Patient was medically fit and no history of any systemic illness was present. The parents were unaffected by hypodontia and among their 5 children, 3 daughters were affected. The two sons were unaffected.

Clinical examination revealed hypekeratinization of palms and eczema of skin on various sites. The prominent extraoral features were periorbital hyperpigmentation, protruberant lips and a prognathic class III appearance. A reduced lower facial height gave the patient an aged appearance (Figure 1). The general oral health was good. The erupted permanent teeth present were UR6, UR3, UL6, UL7, LR6, LR5, LR3, LL6, LL5, LL3. Retained primary teeth were ULb and ULc (Figure 2, 3). The teeth present also exhibited microdontia which is a common and a concurrent finding along with hypodontia. In centric occlusion the patient had contacts on RU6, RL6 on one side and LU6, LL6 on the other side (Figure 4). Radiographic examination showed no unerupted permanent teeth (Figure 5).

Case 2

The second sister was 12 years old. Her mother was concerned that her primary teeth never shed and the permanents did not erupt. Otherwise, the patient was fit. Clinical examination revealed facial symmetry, periorbital hyperpigmentation and eczema on various sites of face and hands. There were no unusual extraoral features except Class III profile (Figure 1). Intraorally, the permanent erupted teeth were UL3, LL3 and LR3. All the primary teeth were retained except ULb, URb and LLc (Figure 2, 3). In occlusion, the posterior teeth were in cross-bite and the anteriors in edge to edge relation (Figure 4). On radiographic examination, the only unerupted permanent teeth was UL3. All other permanent teeth were congenitally missing (Figure 5).

Case 3

The youngest sister was 6 years old. The mother brought her for a routine dental checkup. The patient had no active dental problem and the medical history was insignificant. Clinical examination revealed no extraoral abnormality except for multiple eczemous patches on the face (Figure 1). Intraorally all deciduous teeth were present except URa and their morphology was a little abnormal as they had small conical crowns indicating microdontia (Figure 2,3). The occlusion was Class I in all molars, canine and incisor relationship (Figure 4). On radiographic examination, the unerupted permanent teeth were UL1, UL3, UL6, UR1, UR6 and LR7. All other permanent teeth were congenitally missing (Figure 5).

Treatment

As both the functional and esthetic problem was most severe in the eldest sister (Case 1), an upper and lower overdentures with soft denture base were fabricated for the patient after discussion of all the possible treatment options. Rest vertical dimension was established and the acylic denture teeth set at the determined occlusal vertical dimension (Figure 6). The remaining teeth were rounded off and undercut areas blocked before the wax up procedure. Final dentures were processed using Molloplast-B. The dentures were retentive and patients esthetics was greatly improved, thus providing both functional and psychological benefit to the patient (figure 7).

The patient was kept on follow up to make necessary adjustments and monitor the oral hygiene of the overdenture abutments. The need for continuous recall visits was greatly emphasized. No active treatment was required for the other two sisters. But they were explained about the importance of preserving the remaining deciduous teeth, advised to follow proper oral hygiene methods and kept on follow up.

DISCUSSION

The prevalence of severe hypodontia is very less. It has been reported to be less than 1 %.[11] Usually it is found to be associated with various syndromes. The three sister presented to us with severe hypodontia, not associated with a syndrome. Out of 5 siblings, 3 were found to be affected with hypodontia which suggest a genetic cause. The condition was most severe in the second sister who had only 4 permanent teeth present. Due to several missing teeth and lack to proper alveolar development, the sisters had reduced lower facial height, protruberant lips and a typical Class III appearance, leading to characteristic appearance of old age.

Patients with congenitally missing teeth suffer aesthetic, functional and psychological morbidity to various degree. Along with missing teeth, these patients suffer characteristic changes in teeth, alveolar volume deficiencies and skeletal jaw mal-relationships. Hence the functional and psychosocial impact is more profound in these patients. Early diagnosis and treatment of such patients is very important.

The treatment of hypodontia is often complex and involves a multidisciplinary approach. The choice of a particular treatment option depends on factors like age of the patient, number of missing teeth, adjacent teeth condition and position, amount of bone resorption and medical factors and financial status of patient. The prosthetic rehabilitation of such patients usually involves removable dentures, resin-bonded bridges and implant supported prosthesis. The various treatment options available must be discussed with the patient and parents. They should be made aware of the prognosis, maintenance implications and final appearance of the suggested treatment option.

The treatment options available for the eldest sister was discussed with the patient and the parents. Because of insufficient quantity of alveolar bone and financial limitation of the patient, implant placement was not possible. Provision of overdentures with soft denture base was planned. The soft denture base was advantageous for the patient as it accommodated the ridge irregularities and changes such as excessive resorption, minimal keratinized ridge epithelium and decreased resistance to irritation due to nutritional and physiologic problems. The patient was made aware of

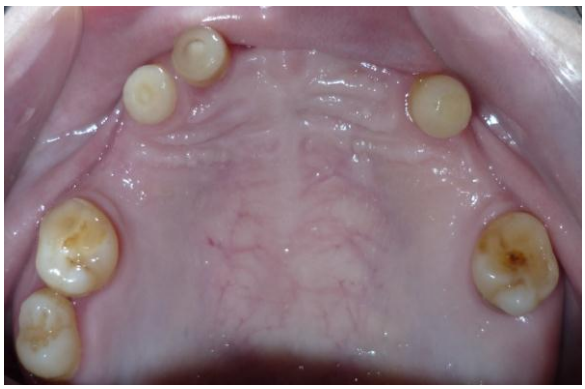
the prognosis, maintenance implications and final appearance of the suggested treatment option. The dentures greatly improved the function and appearance of the patient.

CONCLUSION

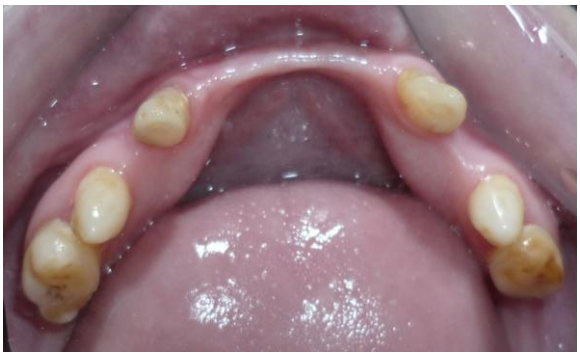
The use of tissue-supported maxillary and mandibular overdenture in the present case can be considered as a good practical alternative that provided a relatively quick, easy, acceptable and economical solution to the functional and esthetic oral rehabilitation.

Case 1:

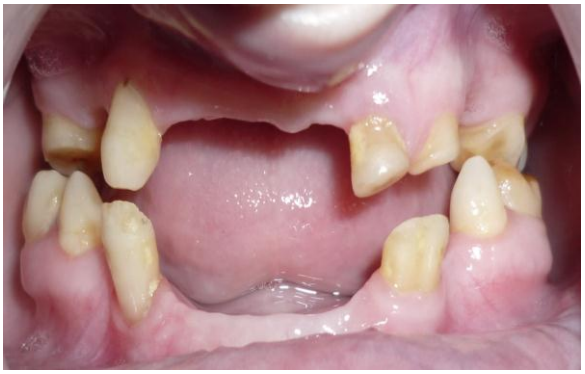
Pretreatment frontal view (figure 1)



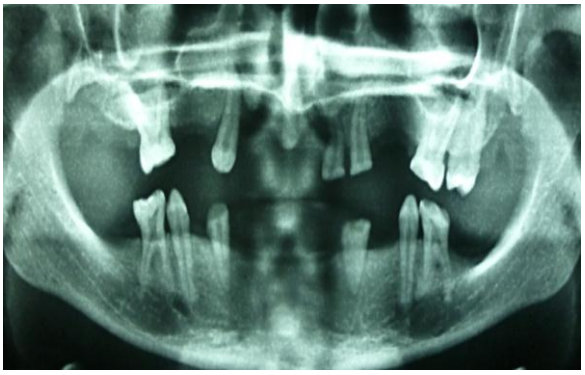
Upper occlusal view (figure 2)



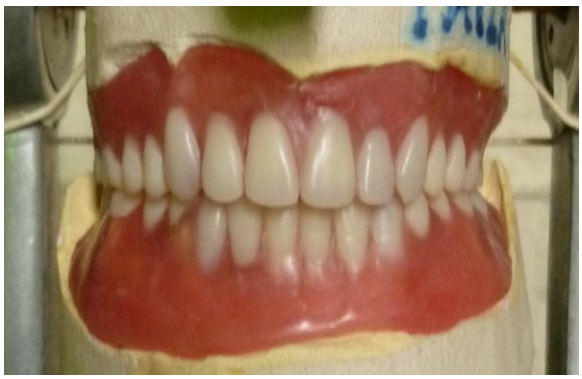
Lower occlusal view (figure 3)



Natural teeth in occlusion (figure 4)



Orthopantomogram (figure 5)

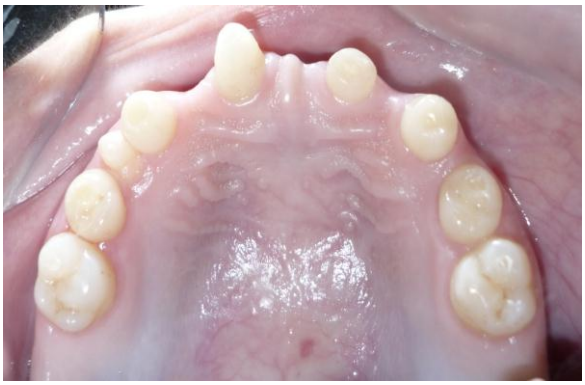


Teeth setup (figure 6)

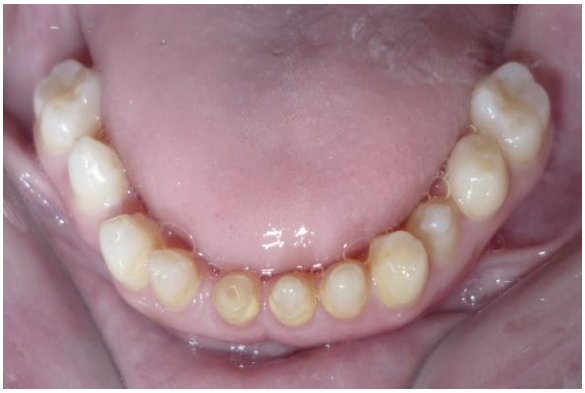
Posttreatment (figure 7)

Case 2

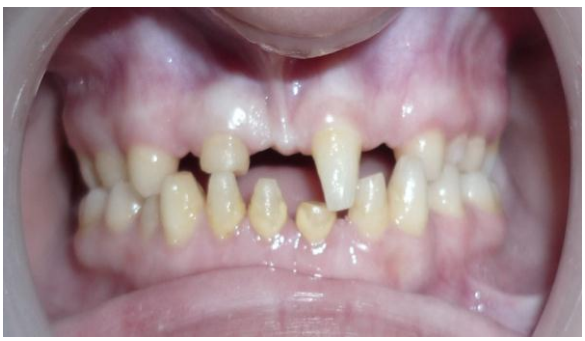
Frontal view (figure 1)



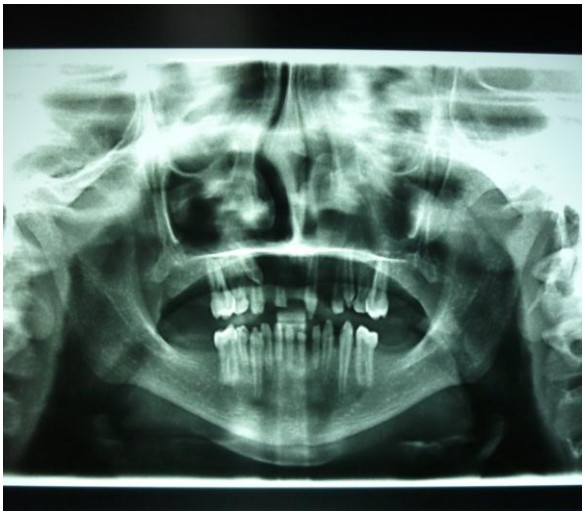
Upper occlusal view (figure 2)



Lower occlusal view (figure 3)

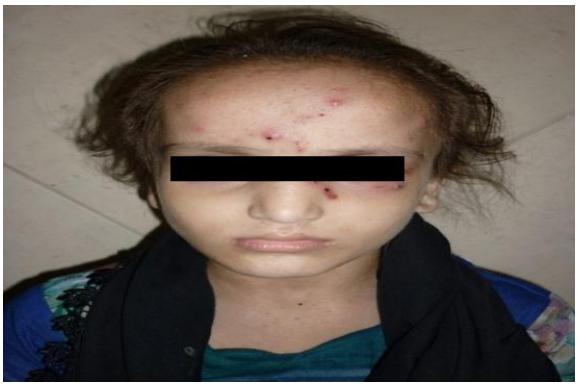


Natural teeth in occlusion (figure 4)

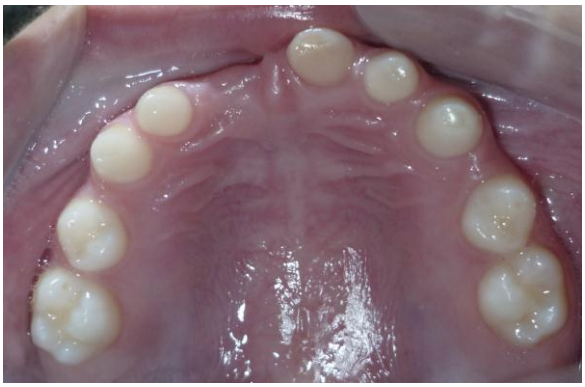


Orthopantomogram (figure 5)

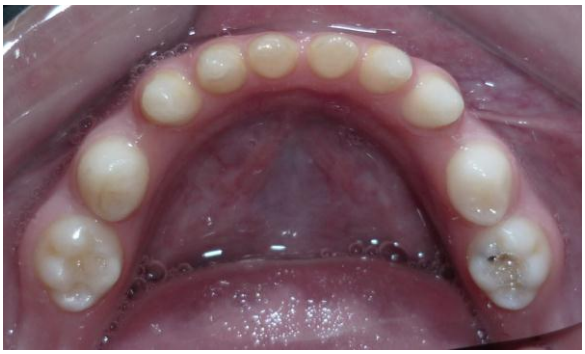
Case 3



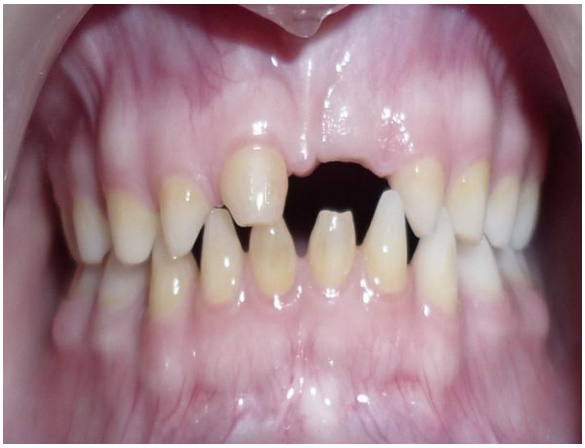
Frontal view (figure 1)



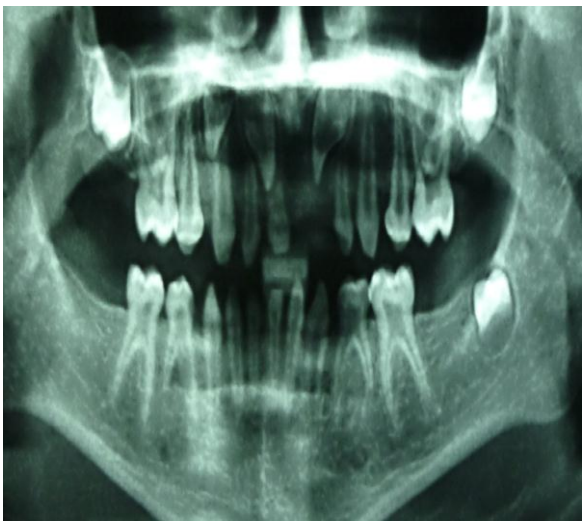
Upper occlusal view (figure 2)



Lower occlusal view (figure 3)



Natural teeth in occlusion (figure 4)



Orthopantomogram (figure 5)

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