ABSTRACT
Congenitally missing teeth (CMT) may present as complete (Anodontia) or partial (Hypodontia or Oligodontia) depending on the number of teeth missing. Although it has been widely reported as a component of a syndrome such as Ectodermal Dysplasia, few reports are available about its sporadic and non-syndromic occurrence. CMT may affect and compromise stomatographic functions such as mastication, speech and esthetics. We report a case of management and rehabilitation of a 5-year-old girl that presented with 8 congenitally missing primary teeth with no evidence of their succedaneous permanent teeth germ. From our investigations, this case was not associated with any known syndrome; hence it is a non-syndromic CMT-oligodontia involving both primary and permanent teeth. We managed both the functional and psychological effect of it on the child and made efforts to preserve the orofacial structures by using Flexible Removable Partial Denture as our treatment modality. It was comfortable for the child, she adapted fast to it, and also the functions, esthetics and her psychological well-being were greatly improved.

Keywords: Congenital, Oligodontia, Tooth Germ, Flexible Denture,

INTRODUCTION
It is rare but not impossible for a tooth to be congenitally missing. Congenitally missing teeth (CMT) is defined as teeth whose germ failed to develop appropriately to allow further differentiation of the dental tissue [1-3]. Congenitally missing teeth, which is also referred to as tooth agenesis or dental aplasia is a developmental disorder but one of the most common dental anomalies which may be sporadic or a part of a hereditary syndrome in occurrence [1-3]. CMT may affect primary or permanent dentition or both and may manifest on the upper or lower jaw. It could be bilateral or unilateral and may affect the posterior or anterior segment of the jaws or both. Dental agenesis may also be total or partial, mild or moderate depending on the number of teeth missing [1, 4]. When it is total (i.e. no teeth) CMT is described as anodontia and this may be a manifestation of an hereditary/developmental anomaly such as ectodermal dysplasia- a disorder where there is disturbance in formation and development of tissues of ectodermal origin probably as a result of mutation during embryonic development leading to defective formation of teeth, hair, skin, nails and sweat glands [4, 5]. Partial CMT depending on the number of teeth missing could be hypodontia or oligodontia. Hypodontia is an anomaly where less than five teeth are congenitally missing while oligodontia is when missing teeth are up to six or more [1, 6-8].

Depending on the number and site of missing teeth, performances of functions like chewing or mastication may become a challenge in CMT. CMT may also cause esthetic embarrassments, lower self-esteem, communication and phonation defects, reduced professional performance, unpleasant facial profile secondary to defective alveolar bone growth and hence the overall quality of life may be affected [2, 4]. Therefore, there is a need to treat the condition so as to restore both functions and the psychological well-being of an affected individual.

The cause of CMT is not yet known. However, some etiological factors have been implicated. These multifactorial etiological factors include both genetically induced and environmental factors. Among the developmental/genetic factors that have been implicated are: ectodermal dysplasia, cleft lip and palate, and Down syndrome. While environmental factors include: trauma, radiations, drugs like chemotherapy, systemic diseases like rickets and syphilis, local inflammations, infections like rubella virus, hormonal disturbance, increased maternal age, low birth weight, multiple births, smoking and exposure of mother to toxins during pregnancy [1, 2, 6, 9-12].

The prevalence of CMT varies from one part of the
world to the other with contradicting prevalence among Africans. While it has been reported that CMT is more common among Africans than any other race in the world, another report suggested a lower prevalence of CMT among Africans [13, 14]. However, there is no strong evidence to support or refute these claims probably as a result of paucity of information and scanty report of CMT disorders in literature from many African countries. Presently, there is no available report in the literature about the prevalence of CMT in Nigeria which is the most populated African country, but in Kenya, a prevalence of 6.30% CMT outside 3rd molars have been reported [15]. Likewise, a prevalence of 10.9% in Iran, 8.50% in Japan, 5.9% in Germany and 8.80% in United States of America have been reported [1, 16, 17]. While CMT disorder has been reported in some studies to have no gender predilection [9, 18], reports from others indicated higher prevalence among female than male [9, 16, 18, 19].

Although unilateral CMT is more widely reported than bilateral, the bilateral CMT involving mostly lateral incisors has been widely reported to be more common in the maxillary jaw than mandible [2, 4]. While the report of some studies showed an equal prevalence of CMT in both jaws, some indicated a higher prevalence in the maxilla than mandible and others reported higher prevalence in the mandible than maxilla [9, 18, 19]. The most widely reported congenitally missing tooth with the highest prevalence in many studies is third molar [19, 20].

The management of CMT is by multi-disciplinary approach involving professionals in the field of dentistry such as general dental practitioners, pediatric dentists, restorative dentists and others such as paediatricians and psychologists just to mention a few. The major concern of dentists is to initiate treatment that will restore masticatory functions, facial profile and the overall esthetic well-being of such patients. Consequently, appropriate treatment modality must be adopted, well planned and structured with adequate consideration for patient’s age and development, the severity of the condition in terms of number of teeth missing and the possible challenges that may arise as a result of treatment of choice. Documented treatment modalities in the literature include; maintenance of primary tooth, placement of implants, provision of a full-full denture, removable partial denture and fixed prosthesis (bridge) [21-24]. Although each of these modalities has its own challenges, it suffices to say that the condition may be easy to manage in an adult than in children with disabilities, and also among the aged.

Report of CMT disorder involving, a set of primary teeth and their permanent successors, bilaterally on both upper and lower jaws in an individual is not readily available in the literature; hence, this case report.

CASE REPORT
A five-year-old girl in the company of her mother presented at the Dental and Maxillofacial Surgery Department of a regional University Teaching Hospital, with a complaint of missing teeth (Fig.1).

According to the mother, the child who is the firstborn in a family of two children has been emotionally disturbed because of the missing teeth. There is no history of such incidence of missing teeth in both the nuclear and extended family. There was no history of consanguineous marriage among the direct family or distance relations of either the father or the mother. According to the mother, she conceived her when she was 30 years of age and that she complied with antenatal attendance. A review of drug intake during pregnancy shows she took Fesolate, Folic acid and Calcium supplements as part of her routine antenatal managements.
She was delivered at 38 weeks through caesarean section at a tertiary health care centre with the child weighing 2.65kg at birth. Since birth, the child’s growth has been normal with no history of any known systemic illness and no history suggestive of intolerance to heat or absence of sweating. There was no history of trauma to the orofacial structure.

Extra-oral examination revealed a healthy-looking girl with no obvious bossing of the frontal of the bone, normal appearance of scalp hair (Fig 1a) and eyebrows with eyelashes that are not loose. The nose is not saddled, upper and lower lips were not protruded, but the philtrum appeared flattened (Fig 1b). The nails were normal in appearance and the skin smooth and fresh without abnormal pigmentation (Fig 1c). Intra-oral examination revealed normal clinical appearance of the tongue, the floor of the mouth, palate, oropharynx and buccal mucosa. The gingivae on the alveolar ridges of the edentulous space on both upper and lower jaws were slightly folded.

**Fig. 2.** Clinical photograph showing congenitally missing primary teeth

![Clinical photograph showing congenitally missing primary teeth](https://www.journal-imab-bg.org)

<table>
<thead>
<tr>
<th>Teeth Present</th>
<th>EDC</th>
<th>CDE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Missing Teeth are</td>
<td>EDC</td>
<td>CDE</td>
</tr>
<tr>
<td>BA</td>
<td>AB</td>
<td></td>
</tr>
<tr>
<td>BA</td>
<td>AB</td>
<td></td>
</tr>
</tbody>
</table>

Histological report of skin biopsy was unremarkable

**Fig. 3.** Micrograph showing histological sections of (a) the skin with normal skin epithelium (b) sweat gland.

![Micrograph showing histological sections of skin](https://www.journal-imab-bg.org)

Histological section of skin tissue harvested showed normal anatomy (Fig. 3). Radiographic investigations revealed congenitantly missing tooth germs of upper and lower primary incisors, their permanent successors, and mandibular second and third molars (Fig. 4). Rehabilitation using Flexible Removable Partial Denture (FRPD) was the treatment modality adopted in the management of this case. After explaining this option to the parents, we obtained their consent before starting the procedure. The first step taken was oral hygiene measures through scaling and polishing of the standing teeth. Impressions of the upper and lower jaws were then taken with appropriate stock trays using alginate (Figure 5a).

Impressions were cast in stone (Figure 5b) and the jaw models were used for designing the flexible RP. The denture was designed and set up of trimmed adult teeth done (Figure 6).

**Fig. 4.** Orthopantomographic view of the patient jaws with congenitally absent tooth germs of upper and lower deciduous incisors and their permanent successors, and mandibular second and third molars.

![Orthopantomographic view of the patient jaws](https://www.journal-imab-bg.org)

**Fig. 5a.** Pictures of upper and lower impression taken in alginate.

![Pictures of upper and lower impression taken in alginate](https://www.journal-imab-bg.org)
Fig. 5b. Stone model on which dentures were fabricated.

Fig. 6. Wax pattern of the removable partial denture framework.

Fig. 7. Dentures on delivery to patients.

The trying-in was carried out, and after the mother and child were satisfied with the colour and the sizes of the teeth, relevant laboratory processes were carried out and the definitive FRPD replacing congenitally missing teeth on maxilla and mandible were produced. The material used for the construction of denture base was Biosoft Flexible Resin-a derivative of Nylon. Flexible RPD was delivered to the patient (Figure 7), and both the patient and the mother were trained on how to insert, withdraw and maintain the dentures. The patient was recalled two days post-delivery, and except for mild pains on mastication from a sharp sport on the denture, which warranted minor trimming, the patient was observed to be satisfied. The patient was later recalled after 2 and then after 4 weeks, and we observed she was coping well without any challenges. Her last visit to the clinic was 8 weeks post-delivery of dentures, and we observed she was satisfied with improved facial appearance, masticatory functions, speech and self-esteem. The patient is now presently on a bi-monthly appointment.

DISCUSSION

This is a case of non-syndromic CMT (oligodontia) with 8 primary teeth missing, and absence of tooth germs of permanent successors that will replace 8 missing temporary teeth and those for the upper and lower second and third permanent molars. It is non-syndromic disorder because no other anomaly was found associated with the CMT. It is well documented that for ectodermal dysplasia (ED) which is the commonest cause of syndromic CMT to be diagnosed at least two of the following features must be well established; Abnormal dentition, Trichodysplasia (Abnormal hair), Onchodysplasia (Abnormal nail) and Dyshydrosis (Abnormal or missing sweat gland) [5]. There was no evidence of the presence of any of the last three in this case. The scalp hair was not scanty, eyebrow with eyelashes appeared normal, the fingernails were growing well, and skin biopsy was not suggestive of any abnormality of the sweat gland. Clinical history did not reveal intolerant to heat or absence of sweat. However, genetic screening is a possibility, although this is not readily available in this environment.

We chose to begin transitional treatment at this age (5 years) in order to boost the patient’s self-esteem and psychological well-being, improve functions and facilitate better facial appearance by normalizing perioral muscle and basal bone growth. Although cases of CMT have been managed in the past with fixed or removable prosthesis, implant and implant supported prosthesis, orthodontic movement of teeth or crowning of well rooted primary teeth with no permanent successor. In this case, flexible, removable partial denture was our treatment of choice for rehabilitating this patient because of the following reasons: flexible, removable partial denture is non rigid with lesser possibility of trauma to the periodontium of the adjacent teeth, residual ridges and adjacent soft tissues during functions, insertion and withdrawal; adaptation is better with flexible than rigid removable acrylic dentures; FRPD is lighter compared to the rein-
forced rigid acrylic dentures; it will be easy to adjust as the child passes through the growing phases; it is a means of encouraging good oral hygiene practices at the early childhood stage as it is easy to clean. Therefore, the use of rigid connector which can interfere with the growth of basal bone in a young patient [5] especially when it crosses the midline was avoided in this case. We equally considered the placement of implant supported prosthesis as not being appropriate because the jaw bones are still growing. Although it is well documented that implant can be placed on anterior mandible as early as 7-12 years according to Guidelines of National Foundation for ED [19], the report some studies have suggested placement of implants after maximum bone growth [6, 9, 13]. The reason may be due to the fact that implants do grow and it may, therefore, interfere and impede both the vertical and horizontal growth of jaw bones, it may also be dislocated, become exposed, ankylosed or cause infraoclusion thereby leading to both functional and esthetic problems especially when used with rigid connectors across the midline in fixed prosthesis.

CONCLUSION
This case report focused on the management and rehabilitation of a 5-year-old girl that presented with 8 congenitally missing primary teeth with no evidence of their succedaneous permanent teeth germ. The choice of flexible, removable partial denture which is lighter and more comfortable to wear than a rigid acrylic denture for the rehabilitation of this patient was based on our conviction that this child will find it easy to tolerate better than rigid removable partial dentures. This treatment modality has improved the appearance of the child, masticatory functions, speech and her psychological well-being. The flexible, removable partial denture will also give room for future periodic adjustment without interfering with the continuous and progressive growth of jaw bones before definitive treatment later in life. It will also boost the child oral hygiene awareness, care and practices early in life.

REFERENCES:


DOI: https://doi.org/10.5272/jimab.2019254.2862

Received: 04/02/2019; Published online: 23/12/2019

Corresponding Author:
Dr Agbara Rowland,
Faculty of Dentistry, College of Health Sciences, University of Jos/Department of Dental and Maxillofacial Surgery, Jos University Teaching Hospital, Plateau state, Nigeria.
E-mail: row_prof@yahoo.com