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Case Report

An Adult with Untreated Isolated Cleft Palate Since Birth: A Rare Case Report

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ABSTRACT

Introduction: Orofacial clefts are one of the most common congenital anomalies involving the craniofacial region. An isolated cleft palate is the rarest (33% of all orofacial clefts) among individuals with female predominance. Individuals born with these clefts manifest craniofacial, functional, and psychological anomalies. Sometimes, patients cannot receive the required therapies due to a lack of awareness and lack of medical access or economic factors. Untreated clefts may adversely impact the patient's life both physically and psychologically.

Case Description: This study presents a case report of a 45-year-old male with an untreated cleft palate since birth due to a low socioeconomic background, leading to several oro-dental anomalies, speech defects, and psychological and social challenges.

Results: Extraoral examination revealed mild facial asymmetry, dry and incompetent lips, and underdeveloped philtrum. Intraorally, there was a cleft palate, macroglossia, soft and edematous gingiva with bleeding on probing, gingival recession, and periodontal pockets, along with various dental anomalies. Full mouth orthopantomogram showed discontinuity of the palatal line, collapsed maxillary arch, root stumps, and crowding. The final diagnosis of isolated cleft palate was made. The patient was advised for surgery regarding the cleft palate, dental treatment, speech therapy, and psychological counseling. However, the patient refused all treatments except a few dental aids due to economic difficulties.

Conclusion: The present case report highlights the need for these patients to receive particular concerns. Patients must be motivated to get proper treatment at a suitable stage to prevent future complications. Cleft teams must be advised to initiate some encouraging steps in imparting treatment aids to such patients, especially in rehabilitation therapy, considering their surrounding environment, psychological makeup, socioeconomic status, and motivation. In addition, social groups should also treat these patients with a positive approach, helping them to live healthy and normal lives just like any other individual.

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Introduction

Orofacial clefts are one of the most common congenital anomalies involving the craniofacial region. These include cleft lip (CL), cleft palate (CP), or both. Their incidence varies worldwide, with an estimated 1 in 700 live births. In India, this incidence is 1 in 1000 live births [1]. The human face's development occurs between the fourth and tenth weeks of gestation (WOG) by rapid migration of the neural crest cells from the neural placode. The fusion of the five basic facial prominences as follows: The midline frontonasal and the paired maxillary and mandibular prominences results in the completion of this process. Defective or lack of fusion between these prominences leads to the development of orofacial clefts. Among all the orofacial clefts, isolated CP is the rarest congenital anomaly (nearly 33% of all orofacial clefts), affecting 1 per 10000 newborns worldwide [2]. The primary palate is formed between the fourth and eighth WOG by the rapid expansion of the frontonasal prominence and fusion of the medial nasal prominences. Failure of fusion of these processes results in the formation of the primary CP.

The secondary palate develops between the eighth and twelfth WOG from the fusion of the palatine shelves, which elongate adjacent to the tongue. As the mandible grows, the tongue descends into the oral cavity, allowing the palate shelves to be elevated above the tongue horizontally [3]. Clefting of the secondary palate may arise from failure of the palatal shelves to elevate, adhere, or fuse. People born with orofacial clefts face a variety of challenges, including facial deformity, dental and oral anomalies, hearing, speech, and respiratory defects, malnutrition, delayed growth, and psychological disorders. The treatment of orofacial clefts is best done in the newborn period, considering all the associated functional abnormalities. The palate should be repaired by the age of 18 months or earlier; however, the treatment of other associated defects may reach up to the teen years. Sometimes, patients cannot undergo timely treatment because of a lack of awareness of early management, access to medical facilities, or economic factors. If left untreated, these clefts may further result in various other complications. Literature has reported a few cases where individuals born with CP were treated in the late stages of their lives. Still, the treatment at that stage could not completely compensate for the craniofacial and physiological abnormalities [4-5]. Isolated cleft palate (ICP) occurs predominantly in females. In this report, we represent a rare case of a 45-year-old male with untreated ICP since birth due to his low socioeconomic background, leading to several oro-dental anomalies, a speech defect, and psychological and social challenges.

Case Description

A 45-year-old unmarried male from a low socioeconomic background working as Gardner was reported to have had a hole in his mouth since birth. History dates back to when the patient's parents first noticed an opening in his mouth. The patient had gone to a local dentist for the same problem and was advised to have surgery; however, the patient refused to go through the procedure. The patient did not give any relevant medical and habitual history. All the family members were healthy. He did not have any problems with eating and drinking but had difficulty in speaking and communication. The patient had to put pressure on his tongue while answering the questions. Meanwhile, he was not pronouncing the words clearly, particularly the consonants. He had a nasal tone in his voice. On general examination, the patient was moderately built with low height (4'3"), normal gait, and posture. He was comfortably seated and well-oriented to time, place, and surroundings. Extraoral examination revealed mild facial asymmetry with slight deviation toward the left side (Figure 1). His lips were dry and incompetent, and his philtrum was underdeveloped. Intraorally, the first notable clinical finding was the presence of an opening in the palate extending from the hard palate (HP) up to the soft palate (SP) (Figure 2). A total of 26 teeth were present (11-17, 21-27, 31, 32, 34, 36-38, 42, 44-48).

There were dental caries i.r.t. 12, 36, 37, 38, 46-48 and root stumps i.r.t. 17, 18, and 38. Other findings were as follows: Collapsed maxillary arch, attrition and crowding in both the maxillary and mandibular anterior regions, buccally placed 13 (Figure 3A), rotated teeth i.r.t. 34,44, and 45 (Figure 3B), generalized stains and calculus. There was slight macroglossia, soft and oedematous gingiva with bleeding on probing, generalized gingival recession, and periodontal pockets. Based on clinical examination, a provisional diagnosis of ICP was made. The patient was advised to have a full mouth orthopantomogram (OPG), which showed discontinuity of the palatal line, a collapsed maxillary arch, root stumps i.r.t 17 and 38, and crowding (Figure 4). Clinical and radiographic pictures confirmed the final diagnosis of ICP. As patients with clefts also have to bear psychological and social challenges besides craniofacial and physiological abnormalities, we tried to evaluate any psychological impacts on him. Hesitantly, he disclosed the rejection and ignorance toward him by his social network, peer group, and even some of his relatives. Based on diagnosis and

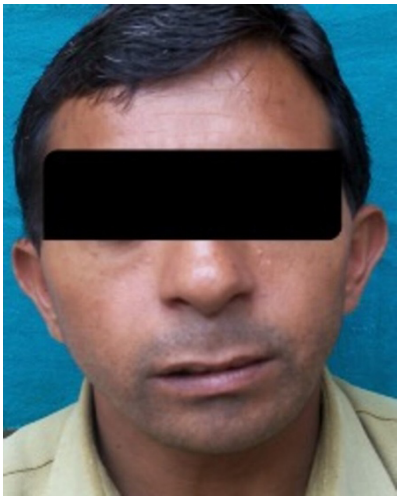

Figure 1. Extraoral photograph showing facial asymmetry

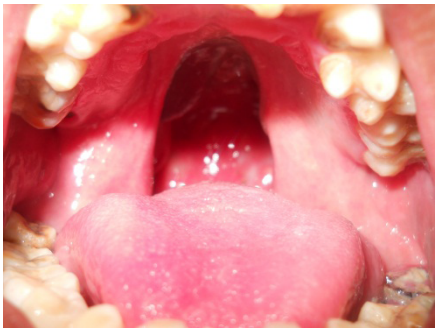

Figure 2. Intraoral photograph showing cleft palate formation

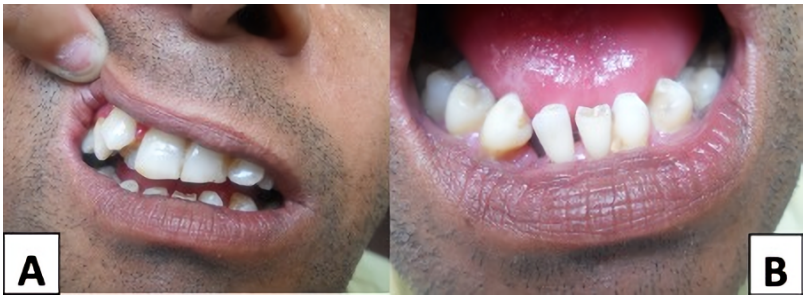

Figure 3. Attrition, crowding, buccally placed 13, rotated 34, 44, and 45

history, we could classify anomalies in this patient as follows: 1) Oro-dental anomalies, 2) Speech defect, 3) Psychological effect, and 4) Social challenges. The patient was explained in detail about the further complications of his disease, and a proper treatment plan was advised, including surgery regarding CP, speech therapy, endodontic treatment for carious teeth, extraction of root stumps, orthodontic aids, oral prophylaxis, and psychological counseling. The patient was ready to extract his root stumps i.r.t. 17 and 38, and restorations done i.r.t. 36 and 37. However, he denied going for all other treatments due to economic difficulties. However, we could provide him with psychological counseling by referring him to the psychology team. The patient was motivated for the follow-up, but it has been a long time since the patient has stopped approaching for further visits, and efforts to reach him have been unsuccessful.

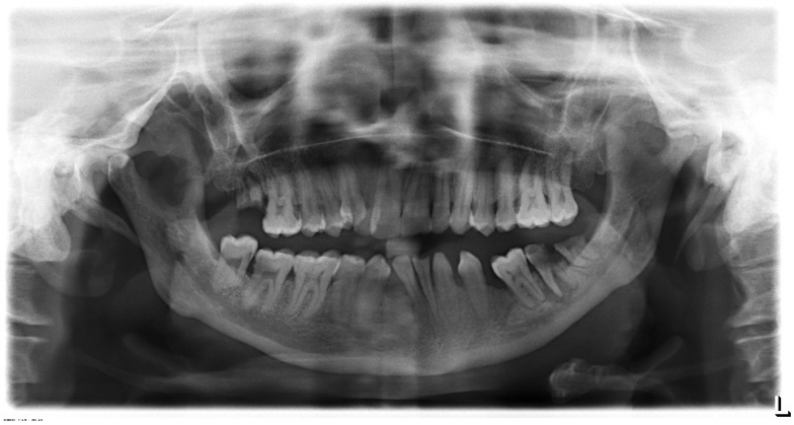

Figure 4. Full-mouth orthopantomogram

JMR

Discussion

Developmental processes play an essential role in the formation of orofacial structures. As discussed, CP is formed due to defective fusion of those processes. ICP is predominantly seen in females. The method of palatine shelf elevation is slightly delayed in females than in males, increasing the risk of CP formation. It is a current hypothesis for the higher frequency of ICP in females [6]. The present case involved a 45-year-old male reflecting a rare occurrence of CP.

The exact etiology of CP is unknown and is suggested to be multifactorial, including hereditary, vascular, lymphatic, and nutritional disturbances, smoking, alcohol, anticonvulsant drugs, radiation exposure, increased maternal age, etc. [7]. In the present case, no such associated factors could be linked to the formation of CP.

Orofacial clefts can be categorized into syndromic and non-syndromic. Individuals born with oral clefts have to take several physiological and psychological challenges depending on the extent of the cleft. One of the immediate and most important concerns for newborn babies is getting adequate nutrition, especially from the mother's milk, to develop a healthy immune system to fight against infections and prevent various chronic conditions in later life. However, due to clefts, the normal reflex mechanism of breast milk suckling is disrupted, especially in CP, resulting in malnutrition in these babies and growth hindrance [8]. Studies have demonstrated that CP children present lower height and growth than healthy children, which may persist as a long-standing effect [9]. Herein, our patient also exhibited a low height (4'3"), which is comparable to any healthy adult of his age group, and the same reasons can be hypothesized for this factor in this case.

These patients manifest with various orofacial anomalies, such as congenitally missing teeth, natal and neonatal teeth, supernumerary teeth, ectopically erupted teeth, enamel hypoplasia, microdontia, macrodontia, rotated, fused or mobile teeth, deep bite, crossbite, protruded maxilla, spacing/crowding, etc. [10]. In the present case, the patient presented with several dental anomalies, such as missing and rotated teeth, dental caries, root stumps, collapsed maxillary arch, crowding, generalized stains and calculus, macroglossia, soft and edematous gingiva with bleeding on probing, generalized gingival recession and periodontal pockets. Poor oral hygiene, the long time required for food clearance, and nasal fluid drainage in the oral cavity are attributed to an increased risk of dental caries in these patients [11]. The lack of adequate density in the alveolar bone of HP to support maxillary teeth results in improper development and exfoliation of teeth, maxillary arch collapse, and crowding. The same factors can be hypothesized for developing oro-dental anomalies in the present case.

Patients with CP frequently exhibit other associated physiological dysfunctions, such as hearing and speech impairments, respiratory issues, and eating and drinking difficulties. Our patient had no such problems except difficulty in speech. Due to breath exiting the nose instead of the mouth and articulation problems resulting from misaligned teeth and abnormal shape of the lip and palate, it is common for cleft children to experience difficulties in speaking clearly and with comprehensible pronunciation [12]. Early palatal surgery can successfully correct speech defects, but delayed treatment adversely affects speech development. Adults with clefts with speech problems during adolescence and adulthood generally do not do as well as patients with clefts who have not experienced significant speech problems [13]. In the present case, the patient has not gone for any treat-

ment for his CP since birth. That might be the reason for his long-standing speech problems.

Besides bearing physiological challenges, patients with orofacial clefts face significant social distress. Literature has revealed that untreated clefts may have psychological impacts on the lives of these patients. Multiple factors, such as unaesthetic appearance, inappropriate speech and communication, inadequate body growth and building, and many other abnormalities, lead to less acceptance and appreciation of cleft individuals by viewers. Sometimes, they are ignored and teased, even by their peers and relatives. Consequently, they may develop some psychological problems, such as behavioral aggressiveness, isolation, and a lack of confidence [14]. More likely, they may even be deprived of an adequate education because they drop out of school early, leading to insufficient future earning potential and living a comfortable life [15]. These patients perceive fewer marriage proposals as resulting in either delayed marriage or being deprived of getting married life. Our patient also revealed the psychological impacts of his untreated disorder on his life. He perceives less liking, rejection, and attention from his peer group, social circle, and even some of his relatives. He does not have a well-known social network. He dropped out of his studies after finishing his third class. Being in his mid-forties, he is still unmarried. He could not get a suitable job due to his speech and communication defects. Presently, he is working as a gardener. Being the only earner in his family, he is struggling to strive for his survival. Many times, he gets depressed and feels isolated, yet he ignores the behavior of others.

The treatment of CP should be initiated at the early newborn stage (approximately 18 months); however, treating the resulting dental and speech problems and associated physiological and psychological implications is usually not fully completed until the late teenage years. New aids of speech testing must be used to evaluate the extent of defects [16]. Literature has reported many cases of adult patients with CP who were treated at the late stage of their lives; however, late treatment results could not compensate for the associated abnormalities completely. Sometimes, patients fail to get proper treatment due to insufficient awareness, medical access, or economic factors. Consequently, delay in treatment leads to further complications impacting the quality of life. Herein, in the present case, the patient had ICP with difficulty in speaking since birth, and he was living with many dental and oral anomalies. Despite being aware of his disorder, he was unable to go for high-cost treatment aids, such as surgical intervention, speech therapy, and other dental procedures, owing to his poor socioeconomic status. He is living life-bearing physical, physiological, and psychological challenges.

Conclusion

Treatment of cleft patients is a difficult task that includes multidisciplinary aids from the newborn stage to the adolescent years, considering all associated craniofacial, functional, and psychological abnormalities. Untreated orofacial clefts may adversely impact the patient's life individually and socially. Treating CP at a late stage demands a different approach and ingenuity. Sometimes, patients cannot get the required therapies due to insufficient awareness, medical access, and economic issues. And the problem is mainly visualized in the developing countries. The present case report of a low-socioeconomic, 45-year-old adult patient with untreated CP since birth bears physiological and psychological challenges that highlight the need for particular concern for such patients. Patients must be educated and motivated to get proper treatment at a suitable stage to prevent future complications. Cleft teams must be advised to initiate some encouraging steps in imparting treatment aids to such patients, especially rehabilitation therapy, considering their surrounding environment, psychological makeup, socioeconomic status, and motivation. Besides this, social groups should also treat these patients with a positive mindset, helping them live a healthy and normal life like any other individual.

Ethical Considerations

Compliance with ethical guidelines

There were no ethical considerations to be considered in this research.

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Authors' contributions

Conceptualization, and writing: Sonia Gupta; Investigation, methodology and project adminisstration: Sonia Gupta and Manveen Kaur Jawanda; Validation: Manveen Kaur Jawanda.

Conflict of interest

The authors declared no conflict of interest.

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