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ORTHODONTIC TREATMENT FOR CLEIDOCRANIAL DYSPLASIA -A REVIEW

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Abstract

Cleidocranial dysplasia (CCD) is a rare autosomal dominant skeletal disorder primarily caused by mutations in the RUNX2 gene, which plays a critical role in osteoblast differentiation and dental development. The condition is characterized by clavicular hypoplasia, delayed closure of cranial sutures, and significant dental anomalies, including delayed exfoliation of primary teeth, multiple impacted permanent and supernumerary teeth, and malocclusion. These dental manifestations present considerable orthodontic challenges, making early diagnosis and a well-coordinated, multidisciplinary treatment essential. This review aims to explore the various orthodontic treatment modalities available for managing CCD, emphasizing both conventional and contemporary approaches. Treatment protocols such as the Toronto-Melbourne and Jerusalem methods are discussed, along with surgical adjuncts and recent advances like digital planning, TADs, and 3D imaging. The article also highlights the importance of individualized treatment planning, long-term follow-up, and patient compliance in achieving successful outcomes.

Introduction

Cleidocranial dysplasia (CCD) is a rare genetic condition primarily affecting the development of bones and teeth, with hallmark features involving the cranial vault, clavicles, maxilla, and midfacial bones. Patients often present with a distinct craniofacial appearance and characteristic dental anomalies, including prolonged retention of deciduous teeth, the presence of multiple supernumerary teeth, and delayed or failed eruption of permanent dentition. Despite these complex manifestations, most individuals do not report functional discomfort in early childhood, and the absence of pain, swelling, or major disability often leads to delayed diagnosis and treatment planning.

As dental deterioration progresses, typically during adolescence, patients begin to experience significant aesthetic and functional impairments, including an edentulous appearance and compromised mastication. This late-onset disability underscores the importance of timely intervention to restore oral function and facial harmony.

Various treatment modalities have been proposed to address the dental challenges in CCD. These include:

- Prosthetic replacement with or without extraction of impacted teeth[1-5]
- Surgical removal of supernumerary teeth with repositioning or transplantation of permanent teeth.[6-8]
- Combined surgical and orthodontic techniques to facilitate the eruption and alignment of natural teeth.[9]

Clinical features

Cleidocranial dysplasia (CCD) presents with a distinct set of craniofacial and dentoalveolar abnormalities, many of which are consistently observed across affected individuals. The severity and expression may vary, but the following clinical features are commonly encountered:

- Retention of Deciduous Teeth A hallmark of CCD is the prolonged retention of primary teeth, often with unresorbed roots, leading to delayed exfoliation and subsequent crowding or impaction of permanent successors.
- Presence of Supernumerary Teeth Multiple supernumerary teeth are a consistent finding. These extra teeth frequently obstruct the eruption path of

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permanent teeth and can alter the normal dental arch form and spacing.

- Delayed Eruption of Permanent Dentition Eruption of permanent teeth is significantly delayed due to diminished eruptive potential. Although the potential is reduced, it is not entirely absent, and spontaneous eruption may still occur in some cases.
- Facial Skeletal Discrepancies Patients often exhibit a decreased vertical dimension of the lower third of the face. A skeletal Class III pattern is frequently observed, primarily due to maxillary hypoplasia combined with upward and forward rotation of the mandible.
- Underdeveloped Alveolar Bone The alveolar ridges show minimal vertical growth, contributing to shallow buccal and lingual sulci. This further compromises the eruption and alignment of teeth and complicates prosthodontic or orthodontic interventions.
- Delayed Eruption of Molars Although anterior teeth often fail to erupt without intervention, the first and second permanent molars usually erupt spontaneously—albeit later than expected—on both arches.
- Delayed Root Development The root development of permanent teeth is significantly delayed, often lagging by approximately three years compared to normal developmental timelines. This can complicate both diagnosis and timing of treatment interventions.[10]

Diagnosis and Radiographic Evaluation

The diagnosis of cleidocranial dysplasia (CCD) is largely based on clinical examination supported by radiographic evidence. Clinically, patients often present with retained deciduous teeth, multiple unerupted permanent teeth, supernumerary teeth, and craniofacial anomalies like frontal bossing, a flat nasal bridge, and underdeveloped maxilla. In many cases, a familial history is present, as CCD is typically inherited in an autosomal dominant pattern due to mutations in the RUNX2 gene.[11]

Radiographically, panoramic imaging is crucial for identifying the multiple unerupted and supernumerary teeth, delayed root development, and the generalized underdevelopment of alveolar bone. Chest and skull radiographs may show clavicular hypoplasia or aplasia and delayed closure of cranial sutures, respectively. Cephalometric analysis often reveals a skeletal Class III pattern due to maxillary hypoplasia and mandibular rotation. In addition, advanced imaging such as cone-beam computed tomography (CBCT) may offer detailed three-dimensional visualization, assisting in precise treatment planning, particularly for orthodontic and surgical interventions.[12]

Cementum analysis in cleidocranial dystosis- Based on studies conducted :

- **Type of cementum in the apical third**
  - In healthy individuals, cellular cementum is consistently present at the apical third of the root in all teeth examined. However, in patients with cleidocranial dysplasia (CCD), this pattern is markedly altered. In a specific CCD case, only one supernumerary tooth exhibited cellular cementum at its apex, while the remaining showed only acellular cementum in the apical region. This deficiency in cellular cementum is thought to play a significant role in the failure of eruption observed in CCD patients.
- **Root resorption at the apex**
  - No signs of apical root resorption were found in the teeth extracted from healthy individuals. In contrast, the teeth from the patient with CCD showed varying levels of resorption at the root apex. Most of the supernumerary teeth exhibited either partial or complete apical root resorption, with only a few remaining unaffected. Similarly, among the permanent teeth, apical resorption was commonly observed, suggesting that apical pathology is considerably more prevalent in teeth affected by CCD
- **Cemento-enamel junction:**
  - Evaluation of the CEJ revealed notable differences between CCD-affected teeth and those from healthy subjects. Each tooth section included two junctions: one on the facial and another on the lingual or palatal side. In the CCD patient's supernumerary teeth, gap-type CEJs were most common followed by edge-to-edge and overlapping cementum
  - In permanent teeth from the CCD patient, gap-type junctions were observed in 100% of cases. Conversely, in healthy individuals, supernumerary teeth showed gap, edge-to-edge, and overlapping types Notably, the average gap length at the CEJ was 209.1 µm in healthy permanent teeth, compared to 109.4 µm and 108 µm in the CCD patient's supernumerary and permanent teeth, respectively.[13]

General principles of orthodontic treatment in cleidocranial dysplasia

Orthodontic management of cleidocranial dysplasia (CCD) is based on timely diagnosis, careful planning, and a coordinated interdisciplinary approach. The condition is often associated with delayed eruption and numerous supernumerary teeth, requiring a combination of surgical exposure and orthodontic traction to bring impacted permanent teeth into the dental arch. Removal of supernumerary teeth is typically necessary to facilitate this process. Fixed appliances are used to align teeth and create adequate space. Given the delayed skeletal maturation in CCD patients, treatment timing must be adjusted accordingly. A prolonged retention phase is crucial due to a high tendency for relapse. Additionally, providing emotional and psychological support plays a vital role in

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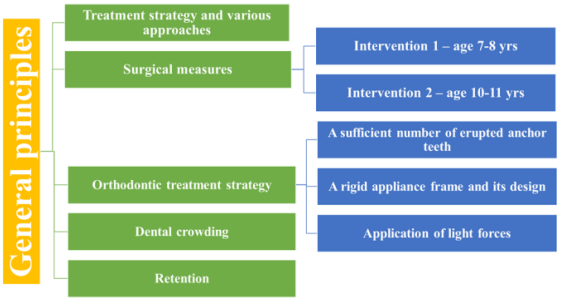
ensuring patient cooperation and overall treatment success.[14]



Treatment strategy of cleidocranial - combined orthodontic and orthognathic treatment

- ***The Toronto Melbourne approach***

The Toronto-Melbourne approach is a staged surgical and orthodontic protocol designed for managing the complex dental issues in cleidocranial dysplasia. It focuses on the early extraction of primary and supernumerary teeth, followed by timed surgical exposure and orthodontic guidance of the permanent teeth.

Treatment Strategy – Toronto-Melbourne Approach

Stage	Age Range	Procedure
INITIAL SURGICAL PHASE	5–6 years	- Extraction of deciduous incisors under general anesthesia. - Removal of supernumerary teeth and overlying bone obstructing permanent teeth.
SECOND SURGICAL PHASE	9–10 years	- Extraction of posterior primary teeth. - Continued removal of any obstructive supernumerary teeth and bone.
RE-EXPOSURE OF INCISORS	After molars erupt	- After first permanent molars erupt and are banded. - Localized re-exposure of permanent incisors and packing of the surgical site. - Placement of attachments on incisors after healing.
PREMOLAR EXPOSURE PHASE	9–12 years	- Surgical exposure of premolars. - Removal of any supernumerary teeth. - Site is packed for healing. - Brackets placed on premolars and canines after recovery.
ORTHODONTIC ALIGNMENT	Later stages	- Although not detailed, a multibanded edgewise appliance is implied to be used for final tooth alignment. [15,16]

- ***The Jerusalem approach***

This approach presents a distinct treatment strategy that is carefully aligned with the underlying abnormalities in dentoalveolar development commonly seen in cleidocranial dysplasia. It is designed based on an understanding of the developmental disruptions that give rise to the syndrome's clinical dental features [17]

Aspect	Description
Core Philosophy	Based on dentoalveolar development and the factors responsible for its disruption in cleidocranial dysplasia.
1. Clinical Features	Focuses on identifying the abnormal characteristics of dentoalveolar structures seen in CCD.
2. Surgical Intervention	Involves precise surgical steps to eliminate supernumerary teeth and remove obstructions to permanent tooth eruption.
3. Orthodontic Planning	Tailors orthodontic strategies to accommodate the unique dental environment and eruption patterns.
4. Aesthetic Consideration	Emphasizes early eruption and alignment of anterior teeth to improve the patient's appearance and psychological well-being.

- ***Belfast Hamburg approach***

The Belfast-Hamburg approach focuses on minimizing the number of surgical interventions by condensing treatment into a single, comprehensive surgical event. Under general anesthesia, all primary and supernumerary teeth are extracted, and the unerupted permanent teeth are fully exposed. A surgical dressing is then placed over the exposed areas to prevent bone regeneration and soft tissue closure, allowing healing by secondary intention. Over time, these surgical packs are regularly changed until conditions allow for the clean and secure bonding of orthodontic brackets to the unerupted teeth. Although some teeth may erupt spontaneously, it is generally not adequate to avoid the use of orthodontic force. Orthodontic appliances are then placed on the erupted teeth, and elastic threads are attached between the bonded brackets of the unerupted teeth and the archwire to guide and encourage proper eruption. [18,19]

Conclusion

Cleidocranial dysplasia is a rare but well-recognized skeletal disorder with distinct craniofacial and dental manifestations. Early diagnosis and a multidisciplinary approach are key to managing the functional and aesthetic challenges associated with this condition. Advances in surgical and orthodontic techniques have significantly improved treatment outcomes, enabling better eruption guidance and long-term dental stability. Individualized treatment planning, considering both skeletal and dental components, is essential for achieving optimal results. With timely intervention and coordinated care, patients with CCD can achieve satisfactory functional and psychosocial outcomes.

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