ISSN: 0975-833X

Available online at http://www.journalcra.com

International Journal of Current Research
Vol. 8, Issue, 07, pp.34164-34167, July, 2016

INTERNATIONAL JOURNAL OF CURRENT RESEARCH

RESEARCH ARTICLE

A NEW ORTHODONTIC-SURGICAL APPROACH IN THE MANAGEMENT OF CLEIDOCRANIAL DYSPLASIA AT THE AGE OF 25-A CASE REPORT

***,[1]Dr. Umapathy Thimmegowda, [1]Dr. Premkishore Kajapuram, [1]Dr. Prasanna Kumar Bhat, [2]Dr. Kumar B. Niwlikar and [3]Dr. Ashwini Chikkanayakanahalli Prabhakar**

[1]Department of Pedodontics and Preventive Dentistry, Rajarajeswari Dental College and Hospital, #14 Ramohalli cross Kumbalgodu, Mysore Road Bangalore 560074
[2]Department of Orthodontics and Dentofacial Orthopedics, Nanded Rural Dental College and Research Centre, Panghari, Nanded, Maharashtra
[3]Department of Oral Pathology and Microbiology, Rajarajeswari Dental College and Hospital, #14 Ramohalli cross, Kumbalgodu, Mysore Road, Bangalore 560074

ARTICLE INFO

Article History:
Received 10th April, 2016
Received in revised form
19th May, 2016
Accepted 23rd June, 2016
Published online 16th July, 2016

Key words:

Supernumerary teeth, Retained teeth, Cleidocranialdysplasia, Orthodontic-surgical regime.

ABSTRACT

The cleidocranial dysplasia is a rare disease which can occur in either sex equally or by autosomal dominant trait. Problems associated with this condition are multiple supernumerary teeth, retained deciduous teeth, delayed eruption, change in shape, impacted teeth and absence of deciduous teeth resorption. For the management of Cleidocranial dyaplasia with many clinical conditions various orthodontic-surgical regimes are discussed in literature. Here we report a rare case of cleidocranial dysplasia in a 25 year old female patient with a new orthodontic- surgical approach which is more adaptive and useful for interdisciplinary management.

Citation: Dr. Umapathy Thimmegowda, Dr. Premkishore Kajapuram, Dr. Prasanna Kumar Bhat, Dr. Kumar B. Niwlikar and Dr. Ashwini Chikkanayakanahalli Prabhakar, 2016. "A new orthodontic-surgical approach in the management of Cleidocranial dysplasia at the age of 25-a case report", *International Journal of Current Research*, 8, (07), 34164-34167.

INTRODUCTION

A female patient aged 25 years reported to private clinic with a chief complaint of delayed eruption of permanent teeth.Patient gave history of uterus cyst and was on medication for the same. Blood investigation was done to rule out abnormalities with thyroid, serum calcium and phosphatase level and found normal. There was no family history of hyperdontia or systemic disease. In the extra-oral physical examination we could notice short height, reduced inter acromial distance, prominent frontal and parietal bones, determining an increase in the cranial perimeter moderately nourished with a concave facial profile (Table/Fig.1). When asked to move the shoulders, she was capable of bringing closer the humeral heads, which characterized the hyper-mobility of the shoulders. The intra oral examination revealed exfoliated lower primary incisors and erupting lower left permanent mandibular central incisor with all other primary teeth retained in both the arches, erupted right and left maxillary permanent first molar, lower left mandibular permanent first molar, atresia of the maxilla and deep palate. (Table/Fig 2) The panoramic radiographic revealed the existence of several teeth which were retained and impacted in maxilla and mandible.

The presence of several impacted permanent teeth, along with multiple supernumerary teeth in the right and left canine and premolar areas and underdeveloped maxillary sinus were seen (Table/Fig. 3). According to the clinical and radiographical findings, the diagnosis of cleidocranial dysplasia was made.

****Corresponding author: Dr. Umapathy Thimmegowda,***
Department of Pedodontics and Preventive Dentistry, Rajarajeswari Dental College and Hospital, #14 Ramohalli cross Kumbalgodu, Mysore Road Bangalore 560074.

Case report

The Cleidocranial Dysplasia (CCD) is a rare disease which occur in elderly individual either in both sex and ethnic group equally or by autosomal dominant trait. There are various clinical findings to be evaluated with this condition like general physical and intra oral. Clinical problems associated with this condition are multiple (Wang *et al*., 2013) Pediatric dentist is the first personto diagnosis this condition and plan interdisciplinary management.

For the management of cleidocranialdyaplasia with many clinical conditions various orthodontic-surgical regimes are discussed in literature. However all the regimes are not appropriate to all individual, it varies from individual to individual. Hence we report a rare case of cleidocranial dysplasia in a 25 year old female patient with a new orthodontic-surgical approach which is more adaptive and useful for interdisciplinary management in this case.

Table/Fig 4. Cleidocranial dysplasia: Orodental anomalies and management options

Anomaly	Management Option	Rationale
Retained deciduous teeth	Removal	Assist eruption of permanent teeth
Supernumerary teeth	Removal	Assist eruption of permanent teeth
Permanent teeth abnormalities	Removal	Construction of removable full/partial dentures (not indicated in childhood)
Unerupted teeth	Retention	Abutments for fixed appliances (not indicated in childhood)
	Surgical exposure	Support for overdenture
	Orthodontic eruption	Function and esthetics and alignment
	Implants	Support overdenture
		Guide impacted teeth into occlusion
	Surgical translocation and/or autotransplantation	Function and esthetics
Malocclusion	Fixed or removable orthodontic appliances	Function and esthetics
Palatal vault narrow-high arched	Expansion with removable orthopedic appliance	Reduce crowding

Table/Fig 5. Cleidocranial dysplasia: Management approaches

Approach		Procedure
Toronto-Melbourne	Age: 5-6	Anterior primary teeth are extracted
	Age: 6-7	Primary incisors are exposed and healing is allowed
		Orthodontic brackets are placed on permanent incisors
		Posterior primary teeth are extracted
Several procedures	Age: 9-10	Permanent bicuspids are exposed
		Surgical removal of supernumerary teeth and healing allowed
	Age: 9-12	Placement of orthodontic brackets on permanent canines and bicuspid teeth
Jerusalem	Age: 10-12	**Phase 1**:
		Anterior primary teeth are extracted
		All supernumerary teeth are extracted
		Permanent incisors are exposed
		Orthodontic attachments are placed on permanent incisors
		Surgical flaps are closed completely
	Age: 13 and older	**Phase 2:**
		Posterior primary teeth are extracted
		Unerupted permanent canines and premolars are exposed
		Orthodontic attachments are bonded
		Surgical flaps are closed completely
Belfast-Hamburg		All primary and supernumerary teeth are removed
		All impacted teeth are surgically exposed
		Surgical packs are placed to prevent healing of bone and soft tissue over teeth
Single procedure		Healing by secondary intention
	Age: not specified	Orthodontic attachments are placed
		Orthodontic appliances placed on fully erupted teeth
		Elastic thread is placed between brackets on unerupted teeth and the archwires
Bronx		**Phase 1**:
		All primary and supernumerary teeth are removed
	Age: not specified	Surgical flaps are closed
Two at most 3 Procedures		**Phase 2:**
		Unerupted permanent teeth are exposed
		Orthodontic brackets are placed
		Surgical flaps are closed and overdenture is placed
		Conventional orthodontic appliances are placed
		Phase 3:
		Leforte osteotomy-orthognathic surgery
		Dental implants are placed

DISCUSSION

Cleidocranial dysplasia was first described in 1765, whereas only in 1898; Marie and Sainton (1898) had described the disease and associated patterns of inheritance. Later, Kallialla (Kallialla and Taskinen, 1962) suggested the genetic mutation as an etiological factor of the disease. In 1962 Lasker and Forlan (1962) had concluded that it was a genetic disease with an autosomal dominant inheritance, skeletal dysplasia with variability (Wang *et al*., 2013). In our case, the patient family members did not report the existence of any clinical characteristic of cleidocranial dysplasia. The radiographic findings showed presence of the deciduous teeth still in the arch, impacted permanent teeth with delayed eruption, although the patient was in her third decade of life. The failure in the eruption can be related with the absence or less amount of cellular cementum in the permanent teeth roots. This was also claimed by Smith, who had observed the absence of cellular cementum in the deciduous and permanent dentition (Counts *et al*., 2001). There are also other hypothesis that explains this fact, as the lack of resorption of the deciduous teeth and subjacent bone and also the presence of a physical barrier, such as supernumerary teeth impacted or by a fibrous connective tissue interposed between the dental follicles and the mucosa. Hence the Pedodontist plays a vital role in the diagnosis of this condition, as well as in the referral and implementation of a therapeutic multidisciplinary planning, aiming the improvement in the life quality of the patients with this condition. The purpose of dental management in patients with CCD is to achieve an optimal functional and cosmetic result by early adulthood. The planning of dental treatment aims in CCD varies from individual to individual and primarily depends on the needs of the patient, the age at diagnosis, and social and economic circumstances (Becker *et al*., 1997).

Nevertheless, the main objectives remain the restoration of craniofacial and dental function together with esthetics (D'Alessandro *et al*., 2010). Depending on the type and severity of anomalies present, a team of orthodontists, maxillofacial surgeons and prosthodontists may be needed to develop an individualized treatment protocol. The options for dental management of craniofacial abnormalities in CCD are summarized in Table/Fig 4 (Roberts *et al*., 2013). The commitment from the entire team at all stages plays a vital role as the treatment continues for a longer duration of time. At the same time, the cooperation from the patient is also very crucial, as serial extractions of impacted and supernumerary teeth may be necessary. Correction of malocclusion may involve surgical repositioning of teeth and the provision of dental prostheses. The dental management of CCD has undergone a metamorphosis from a "wait and observe" approach to more sophisticated and costly methods combining orthodontics and surgery (Farronato *et al*., 2009).

Although there are numerous options, there is a general consensus that the best results are obtained if the condition is diagnosed and treated at an early age. The most popular orthodontic-surgical regimes which are used in the management of CCD are the Toronto-Melbourne, Belfast-Hamburgh, Jerusalem approaches and Bronx approach are summarized in Table/Fig 5 (Roberts *et al*., 2013).

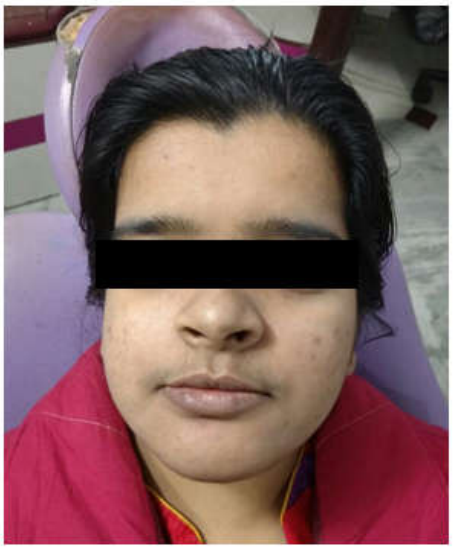

Fig. 1. Anterior view of the patient with cleidocranial dysplasia. Observe the prominence of the frontal and parietal bones, determining the increase of the cranial perimeter

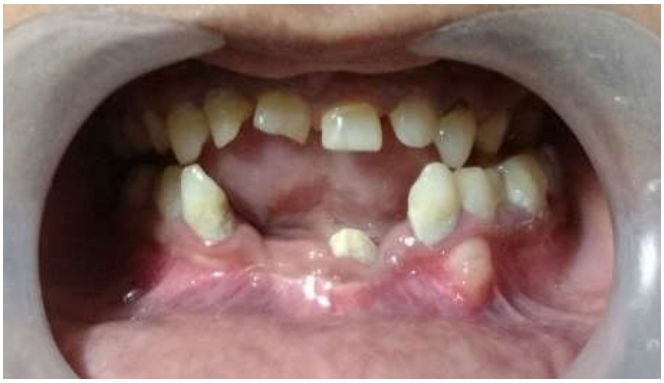

Fig. 2. Intra oral view of the patient with retained deciduous teeth in mouth

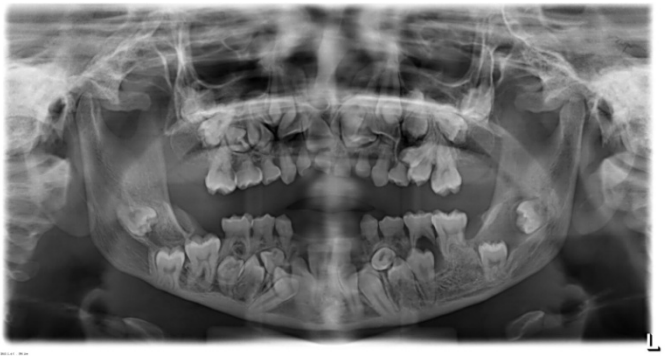

Fig. 3. Panoramic radiography that shows the existence of several dental elements retained and impacted in maxilla and mandible, supernumerary teeth and underdeveloped maxillary sinus

A new modified approach is considered in our case which involves a combination of Toronto-Melbourne, Jerusalem and Bronx approaches. Stage wise management is considered and implied accordingly in our case. This protocol is followed in our case as it is an ideal approach in management of such cases, since management varies from individual to individual.

Stage 1: Firstly it involves bonding and banding of orthodontic attachments for both upper and lower arch including existing primary and permanent teeth which is the main rigid arch wire.

Surgical exposure of the lower right first permanent molar, followed by bonding molar tube for the same. Then uprighting and extrusion of lower right first permanent molar is carried out through orthodontic mechanics by placement of continuous arch wire. Sequence of arch wire used for leveling and alignment are owen 6 Niti, 17x25 Niti, 19x25 Niti, 19x25 Stainless steel wire (SS Wire). This approach reduces the physcological trauma to the patient by disipmacting the individual teeth so it does not impair the function and esthetics of the patient.

Stage 2: After Completion of alignment and leveling in both upper and lower arch it is followed by placement of 19x25 SS wire for both the arches. As the patient is in her third decade of life, removal and surgical exposure of retained deciduous teeth, impacted supernumerary and permanent teeth needs to be carried out according to the sequence of eruption and not according to age. Sequence of surgical removal/exposure of impacted supernumerary teeth are as follows; first lower anteriors, upper anteriors, lower premolars followed by upper premolars (extraction of impacted supernumerary premolars are decided based on bone level, root completion level, position of tooth in the arch), upper and lower canines, second molars, third molars.

After Surgical exposure of teeth according to sequence of eruption, each tooth is brought to occlusal level with the help of elastics tied to the main rigid arch wire. As lower anteriors have no preceeding primary teeth, they are surgically exposed and brought into occlusion with orthodontic attachments, elastics and were included into the main arch wire and the anchorage is reinforced. The same procedure for upper anteriors needs to be done with the removal of retained deciduous teeth, followed by the removal of lower primary first and second deciduous molar and impacted supernumerary premolar. Surgically exposing the impacted lower premolar unilaterally or bilaterally depending on the compliance of the patient needs to be carried out followed by bonding of orthodontic attachment to the impacted teeth. Then teeth are pulled to the occlusal level by using elastics tied to the main arch wire. Once lower premolar comes into occlusion they are included in to the main arch wire and anchorage is reinforced. The same procedure is followed for upper premolars. Second molars are up righted with implant supported uprighting spring made up of 17x25 TMA wire. Third molars which are impacted are advised for extraction because they are not in favorable.

After completing orthodontic treatment, contouring of malformed teeth will be carried out with composites and crowns for esthetic purpose as prosthodontic management. Permanent lingual retention will be placed in both upper and lower arches. The new modified treatment approach is an ideal approachin the present case as it provides better anchorage system to dismpact the impacted teeth.

Conclusion

Various orthodontic- surgical regimes are discussed for the management of CCD but ideal approach are those which adapts to clinical condition of each individual and which improves the function, esthetics and benefits more for the patient. So the new modified approach is adequate enough to treat this patient more efficiently. So this can be implied to other patients with same clinical situations as in need.

REFERENCES

Becker A, Lustman J, Shteyer A. 1997. Cleido- cranial dysplasia: Part 1-General principles of the orthodontic and surgical treatment modality. *American Journal of Orthodontics and Dentofacial Orthopedics*., 111:28-33. http://dx.doi.org/10.1016/S0889-5406 (97)70298-1.

Counts AL, Rohrer MD, Prasad H, Bolen P. 2001. An assessment of root cementum in cleidocranial dysplasia. *The Angle Orthodontist*, 71, 293-298.

D'Alessandro G, Tagariello T, Piana G. 2010. Craniofacial changes and treatment of the stomatognathic system in subjects with cleidocranial dysplasia. *Eur J Paediatr Dent*, 11:39-43.

Farronato G, Maspero C, Farronato D, Gioventù S. 2009. Orthodontic treatment in a patient with cleidocranial dysostosis. *Angle Orthod*., 79:178-85.

Forlan M. 1962. Cleidocranialdysostosis. *Am. J. Med*., 33(2):792- 9.

Kallialla E, Taskinen PJ. 1962. Cleidocranialdysostosis. *Oral Surg*., 15(8):808-22.

Marie, P. and Sainton P. 1898. Sur la dysostosecleido-craniennehereditaire. Revista de Neurología, 6:835-838.

Roberts T, Stephen L, Beighton P. 2013. Cleidocranial dysplasia: a review of the dental, historical, and practical implications with an overview of the South African experience, *Oral Surg Oral Med Oral Pathol Oral Radiol*., 115(1): 46-55.

Wang S, Zhang S, Wang Y, Chen Y, Zhou L. 2013. Cleidocranial dysplasia syndrome: clinical characteristics and mutation study of a Chinese family, *Int J Clin Exp Med*., 6(10):900-907.

****