

ÇOK SAYIDA SÜREMEMİŞ DİŞLERİN ORTODONTİK VE CERRAHİ TEDAVİLERİ (Bir cleidocranial dysplasia vakası nedeniyle)

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ÖZET: ÇOK SAYIDA SÜREMEMİŞ DİŞLERİN ORTODONTİK VE CERRAHİ TEDAVİLERİ (Bir cleidocranial dysplasia vakası nedeniyle) Bu vaka raporunda, dişlerinde sürme bozukluğu olan ve çok sayıda supernumerer dişe sahip olan cleidocranial displazili bir olguya uygulanan cerrahi ve ortodontik yaklaşımlar sunuldu. Eskiden bu tip vakalar sadece protetik yaklaşımlarla tedavi edilirdi. Ancak tedavi alternatifleri zamanla gelişti. Yinede bu tür vakaların tedavileri oldukça güç, uzun ve interdisipliner çalışmalar gerektirir. Bu vaka raporunda da interdisipliner yaklaşımlarla sürememiş olan dişler sürdürülerek sonuçta okluzyonda başarılı bir fonksiyon ve estetik sağlandı.

Anahtar Kelimeler: Sürememiş dişler, artı dişler, cleidocranial displazi

SUMMARY: ORTHODONTIC AND SURGICAL TREATMENT OF MULTIPLE UNERUPTED TEETH (Report of a Cleidocranial Dysplasia Case) In this case report, a cleidocranial dysplasia case having failure of the teeth to erupt and multiple unerupted supernumerary teeth was introduced and orthodontic and surgical treatment was explained. This type of cases was only treated by prosthetic approach in the past. Recently, several treatment alternatives has been developed. However, the treatment of this type of cases is still difficult and requires long and interdisciplinary work. This case report described unerupted permanent teeth, to move them into a satisfactory functional and aesthetic position with interdisciplinary management.

Key words: Unerupted teeth, supernumerary teeth, cleidocranial dysplasia

INTRODUCTION

In order for a permanent tooth to erupt the overlying bone and primary tooth roots must resorb and the tooth must make its way through the gingiva. Supernumerary teeth, sclerotic bone and heavy fibrous gingiva can also interfere with eruption. Multiple supernumerary teeth contribute on element of mechanical interference (1). Supernumerary teeth may delay eruption as well as diastemas, displacement or rotation of the adjacent permanent teeth (2-4). They may also lead to the deve-

lopment of dentigerous or primordial cysts and root resorption of the adjacent teeth (5).

Although the aetiology of the supernumerary teeth is still unknown, several factors such as dichotomy of the tooth bud (6, 7), genetics (8, 9) and various syndromes (3,10-13) are considered as aetiological factors in previous studies. One of the syndrome with supernumerary teeth is cleidocranial dysplasia (CCD) which is an autosomal dominant or rarely autosomal recessive and generalized skeletal disorder characterized by aplasia or hypoplasia of the clavicles, enlarged calvaria with frontal bossing, abnormalities of the cranial base, multiple wormian bones, distal phalanges with abnormally pointed tufts, hypoplasia of the pelvis and many other abnormalities (13, 14-19). The most characteristic clinical sign of the cleidocranial dysplasia is the eruption of the permanent dentition. The primary dentition seems to develop in relatively normal way while the permanent dentition is severely disturbed with predisposition for multiple supernumerary teeth, failure of eruption, ectopia and abnormal tooth morphology, especially involving the roots (15, 20-23).

CCD occurs with equal frequency in both sexes (15). The defect was thought to involve only bones of intramembranous origin but it is now recognized that bones of intracartilaginous origin are also involved.

In this case report, a cleidocranial dysplasia case having failure of the teeth to erupt and 8 unerupted supernumerary teeth is introduced and orthodontic and surgical treatment is explained.

Case Report

A 16 year old Turkish girl N.Y were referred to the Faculty of Dentistry University of Gazi because of uneruption of the maxillary anterior permanent teeth. When she had received routine clinic and radiological examination it was observed that she had anterior openbite heavy fibrous gingiva and still retained anterior primary teeth in the maxilla, primary canines in the mandibula and she had deep caries of maxillary and mandibular first primary molars. The palate was high and narrowed slightly. A class III malocclusion was present

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(Fig.1a-1e). Orthopantomograph showed she had unerupted permanent teeth besides 8 supernumerary teeth with three in the maxilla and five in the mandibula. Two of the maxillary supernumerary teeth were seen between central incisors and the third was lying between the right canine and premolar regions. Two of the mandibular supernumerary teeth in the shape of the laterals were found between lateral and canine on each side. One supernumerary tooth in the shape of the premolar were lying between the left premolars and the other two supernumerary teeth were seen between left first molar and first premolar. Also on this region, there was a dentigerous cyst (Fig. 2a-2c). As these results indicates the CCD syndrome, complete radiographic work up was done. The clavicles vell whole but acromial portion were hypoplastic (Fig.2d). Sacroiliac joints were slightly wide (Fig.2e).The skull graphic represented brachcephalic indices, poor visualization of the posterior cranial base, flexion of the clivus and short anterior cranial base (Fig.2f). These findings were consistent with the diagnosis of the cleidocranial dysplasia. The family history was significant as the family also had supernumerary teeth (Fig.3). However it was not possible to comment on displasia since radiological examination of her family was not at our disposal.

The decision was made to operate dentigerous cysts and to extract the teeth on the cystic region and retained primary teeth. The heavy fibrous gingiva on the anterior maxillary region was removed and supernumerary teeth were extracted step by step. Spontaneous eruption of the unerupted permanent teeth was expected. Those events lasted in two years. Active orthodontic treatment was started at the end of the second year (Fig.4). Class I occlusion with anterior openbite was obtained at the end of 1 year and 9 months of orthodontic treatment (Fig.5a,5b). The remaining anterior openbite was eliminated by anterior subapical osteotomy (Fig.6a,6b). Ideal occlusion was achieved by the application of post-surgical orthodontic treatment (Fig.7a-7g). There was a bolton discrepancy resulted from mandibular anterior teeth. Therefore, the size of upper canines were restored aesthetically. No relapse was observed after one year (Fig.8a-8g).

THE CURRENT MODE OF THERAPY

First surgical approach:

Step by step to remove all deciduous teeth, supernumerary teeth and cysts and to expose all unerupted teeth simultaneously.

Orthodontic Approach:

Upper Jaw: After spontaneous eruption, bond attachments was applied to the upper jaw (Fig.4) and the following implementations were carried out.:

*0.18 x 0.25 slot Edgewise Appliance (75431 I 13457); closing diastema and canine distalization with elastics on sectinal arches + cervical headgear for anchorage (4 months).

* 0.014" SS multiloop arch (1.5 months), 0.014" SS straight arch + Second molar distalization for CI I molar relation (2 months).

*0.016" SS straight arch + bilateral cross elastics to the second molars (1.5 months).

* Palatal expansion with Haas appliance (1 months)

*0.018" NiTi arches and retention of palatal expansion with a removable appliances (3 months).

*0.016x 0.022 " SS arch (2 months), 0.017 x 0.022" SS arch (1.5 months).

Lower Jaw:

*0.018x0.025 slot Edgewise Appliance (754321 I 123457).

* 0.014" SS multiloop arch (*2 months), 0.016 " SS multiloop arch (1.5 months).

* 0.016" SS straight arch (1.5 months), 0.018 straight arch (3 months).

* 0.016 x 0.022" SS arch (2 months), 0.017 x 0.22" SS arch (1.5 months).

Total duration of presurgery orthodontic treatment : 1 year and 9 months (Fig.5a,5b).

Second Surgical Approach:

Anterior subapical osteotomy in the mandible, commonly has been used to close anterior openbite (24, 25) was allied to this case. After the vestibular incision made beginning in the first molar region and the carried around to the opposite site. Mucoperiosteal flap reflection was carried out. The neurovascular bundle removed from its canal was protected. Using a bur, a vertical osteotomy was carried through the alveolar bone in the planned area, just distal to the second premolars and a horizontal osteotomy 4 - 5 mm. below the root apices connected the vertical osteotomy sites. After the dentoalveolar segment was freed; it was elevated and repositioned, maxillomandibular fixation was established in the planned occlusion; an allogeneic bone graft was wedged between the repositioned segment and intact mandible and fixed with two miniplates. The wound was closed with 4x0 nonresorbable monofilament suture.

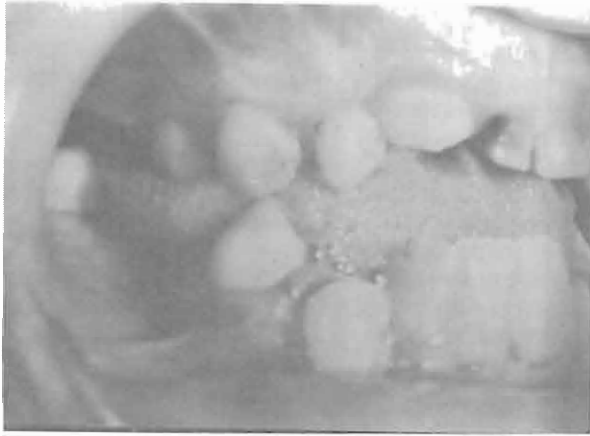


Fig. 1a



Fig. 1b



Fig. 1c

Fig.1a,1b,1c, Pretreatment intraoral and facial photographs of N.Y.



Fig. 1d

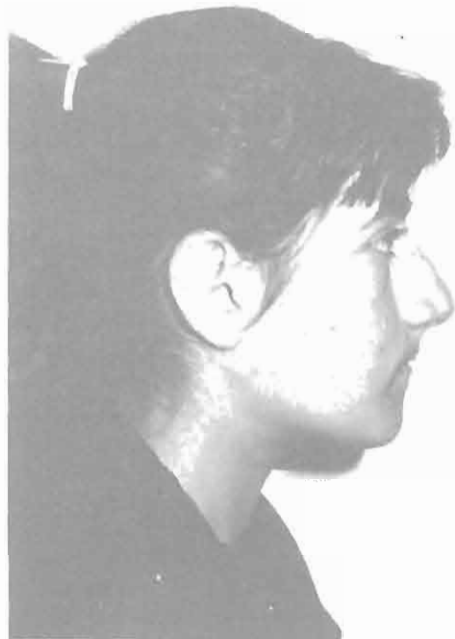


Fig. 1e

Fig.1d,1e |Pretreatment intraoral and facial photographs of N.Y.

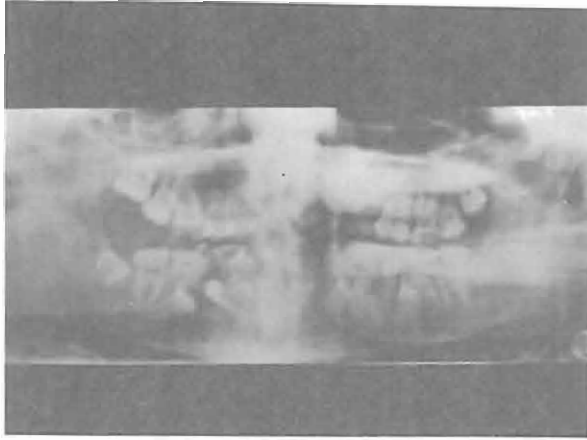


Fig. 2a

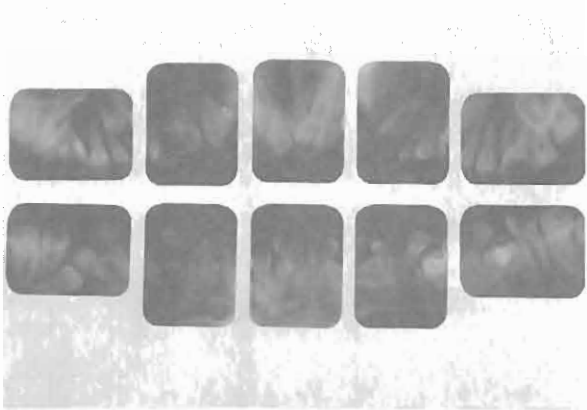


Fig. 2b



Fig. 2c

Fig. 2a,2b,2c Pretreatment radiographs of N.Y.



Fig. 2d



Fig. 2e

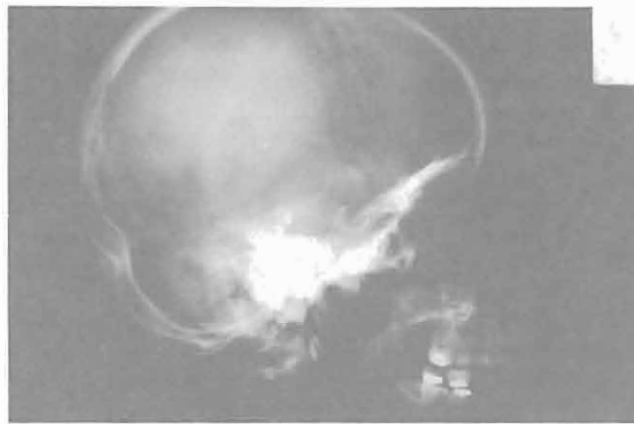


Fig. 2f

Fig. 2d,2e, 2f Pretreatment radiographs of N.Y.

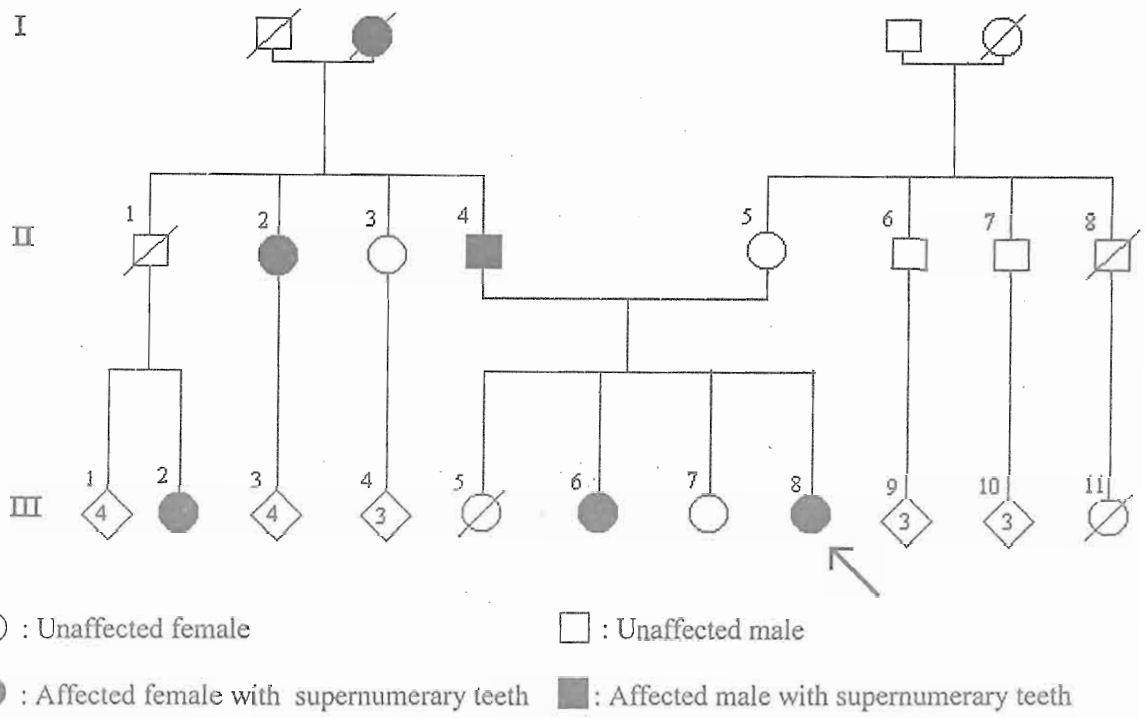


Fig. 3 Pedigree of case N.Y

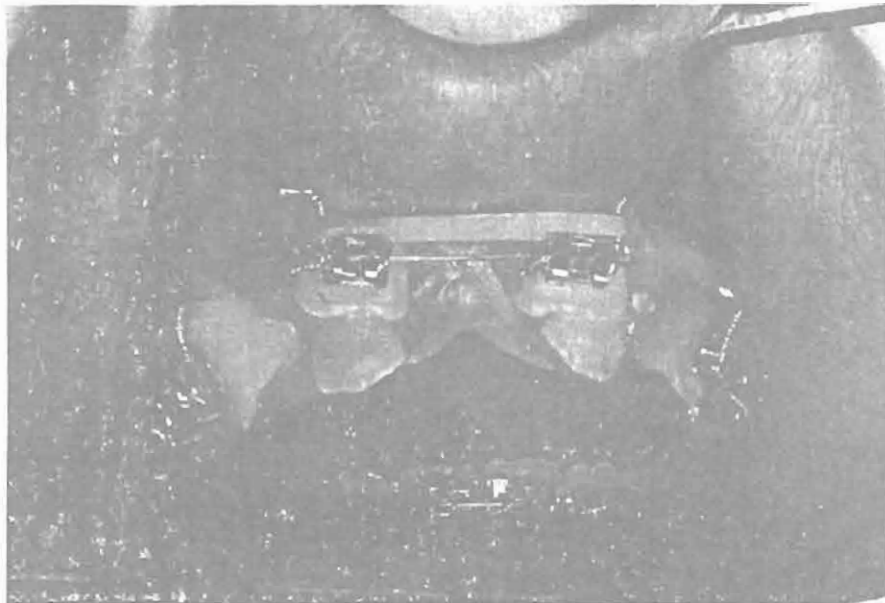


Fig.4 The phase of spontaneous eruption and the first orthodontic treatment.

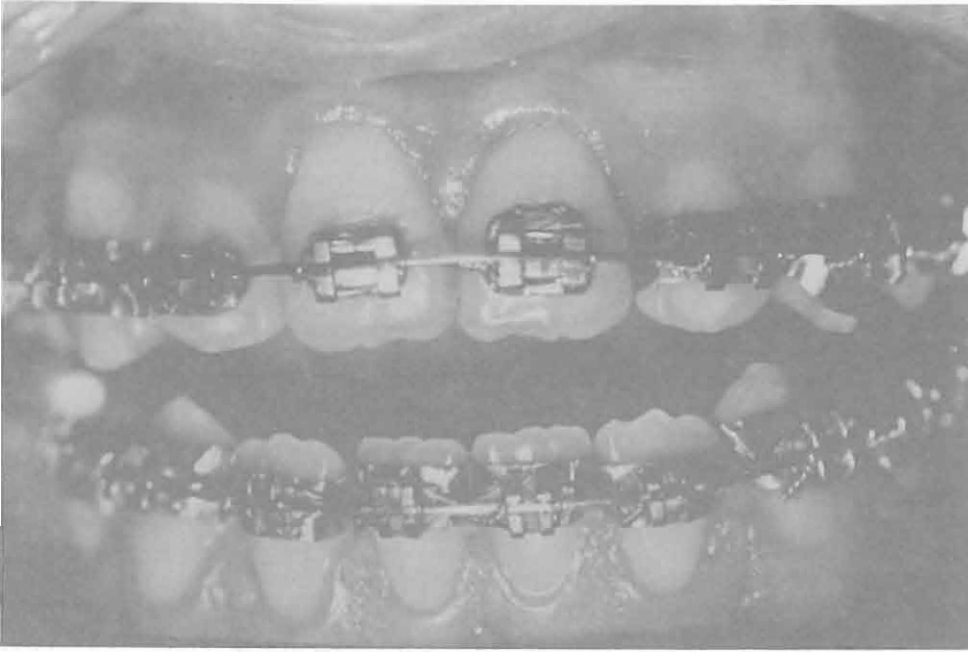


Fig. 5a



Fig. 5b

Fig. 5a,5b Presurgical intraoral and radiographic views.

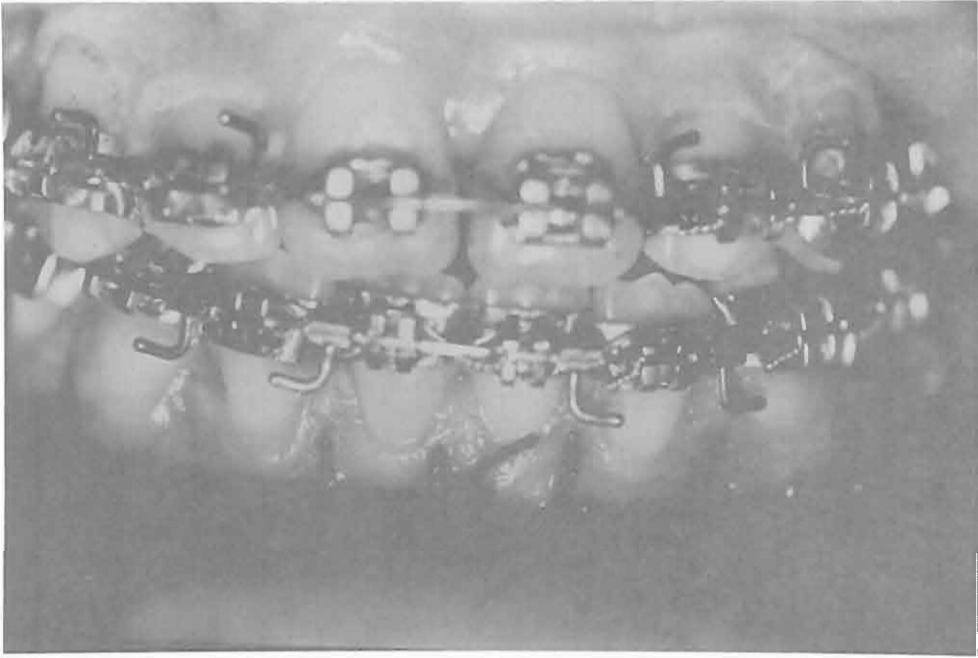


Fig. 6a



Fig. 6b

Fig. 6a,6b Postsurgical intraoral and radiographic views.



Fig. 7a



Fig. 7b



Fig. 7c

Fig. 7a,7b,7c Intraoral, facial and radiographic views after orthodontic treatment.



Fig. 1d



Fig. 1e

Fig. 7d,7e Intraoral, facial and radiographic views after orthodontic treatment.



Fig. 7f

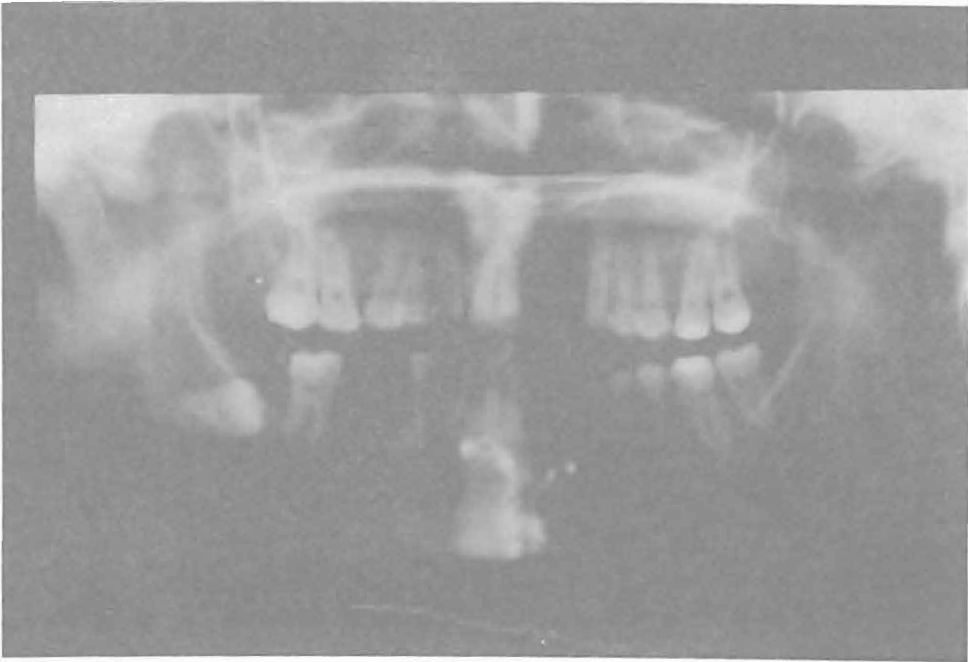


Fig. 7g

Fig. 7f,7g Intraoral, facial and radiographic views after orthodontic treatment.



Fig. 8a



Fig. 8b



Fig. 8c

Fig.8a,8b,8c Intraoral and radiographic views in retention.



Fig. 8d



Fig. 8e

Fig.8d,8e Intraoral and radiographic views in retention.



Fig. 8f

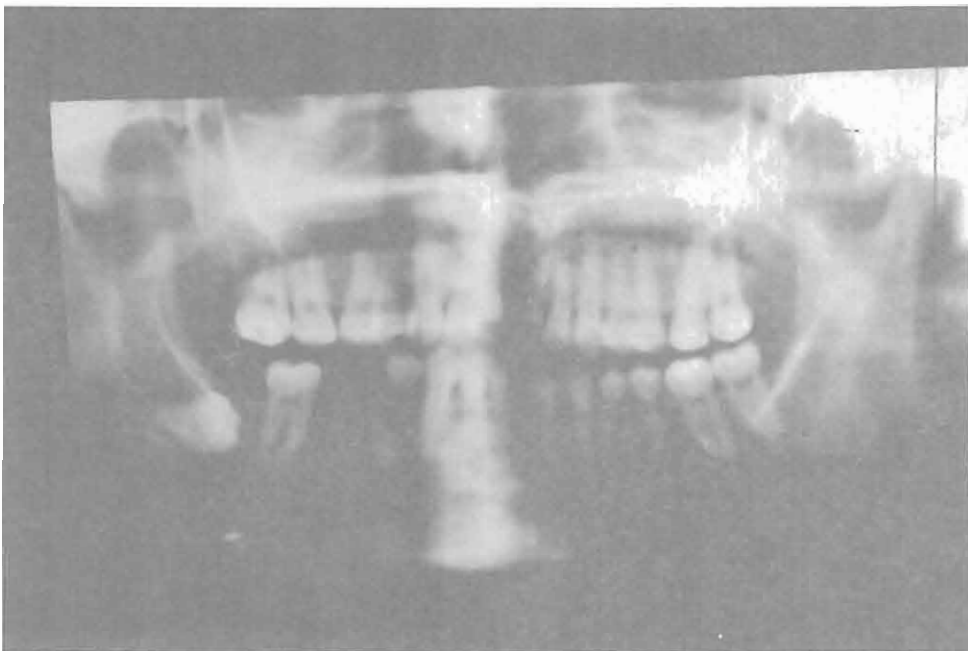


Fig. 8g

Fig.8f,8g Intraoral and radiographic views in retention.

Post Surgical- Orthodontic Approach:

During the operation the front part between both premolars of the 0.17x 0.22 SS arc wire existed in the lower jaw was cut. Another arc wire with the same thickness was soldered into the cut wire so that it could reach posterior teeth passively. Following the release of intermaxillary fixation, boxes elastics were attached to the anterior region. After one month, since the brackets adjacent to the osteotomy are not aligned, the lower archwire, 0.16x 0.22 TMA with T loop, was placed at the osteotomy region. The teeth adjacent to the osteotomy are ligated together as tightly as possible to close any residual space or to prevent reopening of any space at this site. The archwire with T loop was used by the patient for two months. Boxes elastics were also used during nights at the same time period. At the end of this period, the archwire with no loops was applied for torque and rotation. Six months after the operation, brackets were removed and hawley appliances were applied to the upper and lower jaws (Fig.7a-7g). The healing was uneventful and the miniplates were removed after 12 months follow-up period (Fig.8a-8g).

DISCUSSION

The lack of eruption in cleidocranial dysplasia (CCD) is due to failure of the overlying bone to resorb (1,21). Histologic examination of the alveolar bone removed from affected patients showed abnormally dense trabeculation with multiple reversal lines, indicating incomplete resorption (21,22). But when unerupted teeth are uncovered, they show a normal eruption pattern. Mechanical interference resulting from impacted supernumerary teeth is also present in CCD.

In this case; there were multiple supernumerary teeth which were mechanical interference and, sclerotic bone and heavy fibrous gingiva. Although CCD is a generalized skeletal dysplasia leading to multiple abnormalities (19), the major concern of the patients is most often the oral / dental disorders.

It is reported by Feldman et al (26) that 20-40 percent of CCD cases has shown new mutations. In this case; Intraoral symptoms of the CCD were typical and also slightly there were the other typical skeletal symptoms. The family of the patient had supernumerary teeth. CCD are inherited as an autosomal dominant trait. The pedigree shows the typical features of autosomal dominant inheritance of the supernumerary teeth (Fig.3). In generation and some persons although have affected parents are normal which could be explained by failure of penetrance. Penetrance is the probability that a gene will have any expression at all (27). It was not possible to reach a conclusion of the CCD in pedigree since a

detailed radiographic analysis of the family of the patient was not at our hand.

Various modes of the treatment have been suggested to manage this problem (28). In general, the extraction of supernumerary teeth and over-retained primary teeth; surgical exposure of unerupted teeth (13) and orthodontic treatment is carried out (29).

According to the Becker et al (30); surgical interventions are governed by the appropriate root developments. Initial efforts toward bringing anterior teeth into the mouth early for the patient's self-image.

If a full complement of the teeth cannot be brought into good occlusion, prostheses will be used to replace missing teeth. If an unfavourable relation of the maxilla and mandible develops, an osteotomy will be considered (14, 30-32).

In this case; adequate surgical intervention at the appropriate time followed by orthodontic management resulted in a satisfactory occlusion.

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