OBLIQUE FACIAL CLEFT AND SUPERNUMERARY TEETH: A CASE REPORT

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Incidence of facial oblique cleft is not precisely estimated ,it was reported as 0.25% [1] and in some investigation as 3.1%[4] of all facial clefts ,it is evident that this is a very rare condition.

This cleft is classified into 3 types : naso-ocular, medial oro-ocular, lateral oro-ocular clefts.

The main reason of this cleft is generally explained as failed fusion of mesoderm in embryonic period (defect in fusion of maxillary process and lateral nasal process is the developmental period [2]), however lateral oro-ocular cleft type and some types of naso-ocular cannot be explained by interruption of fusion of mesoderm in embryonic facial process, so amniotic bands are reported to be the main cause of these clefts in some investigations [3].

The oblique clefts were frequently associated with other types of clefts and/or more distant anomalies such as scoliosis, microcephaly, microphthalmia and corneal opacity[5] . There are several theories about the cause of these cleft , no obvious genetic chromosomal or environmental factors were found to explain their etiology[6]. In some rare cases taratogens were said to be a probable cause[7] ,Amniotic bands could possibly be thought to be responsible for the severe malformations .

If amniotic bands are adherent to some regions of the developing embryo ,these clefts , other morphogenesis Including constriction rings, aplasia cutis, and other unexplained facial clefts occur. If an amniotic band is interposed between adjacent facial processes , it prevents their fusion ,and cause a lip, palate, and other clefts such as oblique facial cleft. [8]

Oblique facial clefts cause by amniotic bands have been reported by many investigators [9,10,11,12]

The lacrimal apparatus was always involved to some degree with the exception of a few incomplete forms. In the alveolar arch the cleft seemed to be situated between the medial incisor and the canine.[6]

Presence of supernumerary teeth in these kind of clefts are so rarely reported.

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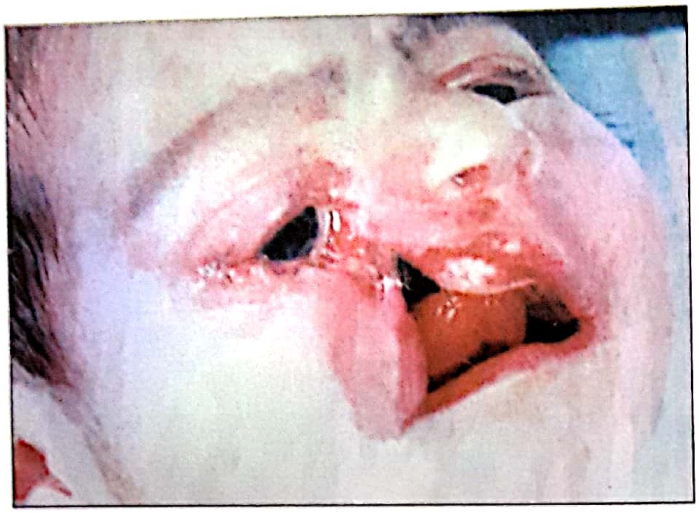

Fig 1a.

Fig 1b.

WHAT IS AMNIOTIC BAND SYNDROME?

Epidemiology

Amniotic banding affects approximately 1 in every 1200 live births. It is also believed to be the cause of 178 in 10,000 miscarriages. Up to 50% of cases have other congenital anomalies including cleft lip, cleft palate, and clubfoot deformity. Hand and finger anomalies occur in up to 80%.

Features

The constriction of appendages by amniotic bands may result in:

1. Constriction rings around the digits, arms and legs
2. Swelling of the extremities distal to the point of constriction
3. Amputation of digits, arms and legs

A strong relationship between ABS and clubfoot exists. A 31.5% of associated clubfoot deformity and ABS can be correlated with 20% occurring bilaterally. Other abnormalities found with ABS include: clubhands, cleft lip, and/or cleft palate, and hemangioma.

Pathophysiology

ABS occurs when the inner fetal membrane (amnion) ruptures without injury to the outer membrane (chorion). Fibrous bands from the ruptured amnion float in the amniotic fluid and can entangle the fetus, reducing blood supply and causing congenital abnormalities. In some cases a complete "natural" amputation of a digit(s) or limb may occur before birth or the digit(s) or limbs may be necrotic (dead) and require surgical amputation following birth.

Diagnosis

At birth, the characteristic asymmetric amputations and constriction rings are seen.

Treatment

Treatment depends on the deformity found. Plastic and reconstructive surgery is often needed. Physical and occupational therapy may be needed long term.

Case report

A girl was aged 7 year and 7 months was referred to orthodontic department of Tehran university with a chief complaint of malocclusion, her physical development was normal for her age ,no significant developmental problem was found in her physical and mental status.

The patient was undergone reparative plastic surgery for her oblique facial cleft.(fortunately the parents of the child give us the chance to have her photograph when she was so young(fig 1))

The parents do not know the exact time and kind of plastic surgery she had undergone and we don't have the complete medical history of her, but the infantile photograph shows the intensity of the cleft, In this picture (fig 1) oblique facial cleft In right zone of the face which involves upper lip and eye is obvious (near fusion line of maxillary process and lateral nasal process (fig 2)).

In the clinical examination, her face was seemed to be asymmetric , her right eye was located inferior when compared with the left one which maybe contributed to surgical scar tissue or other factors. Labial cleft was satisfactory repaired . But cheek and suborbital defect besides a lot of surgical processes was obvious.

There was no family history of cleft lip and palate, and the mandibular arch was normal.

There were three first deciduous molar and three second deciduous molar and a first permanent molar on the left side of maxillary dental arch.(fig 3)

Panoramic radiograph reveals supernumerary teeth in upper arch. (fig 4)

Discussion

This patient have two rare characteristic ,first of all is the

Fig 2.

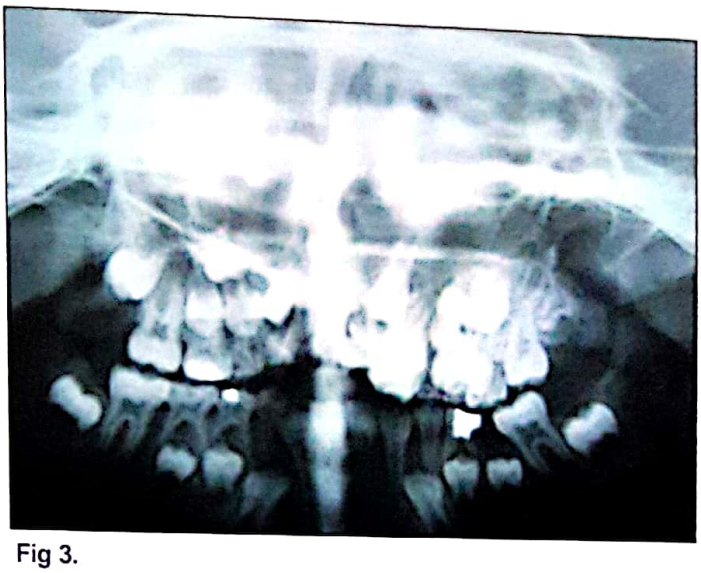

Fig 3.

Fig 4.

presence of seven teeth in upper left dental arch and the other is the presence of these teeth in the opposite side of cleft .

frequency of supernumerary teeth in this kind of cleft is so rare and if this happens the supernumerary teeth usually erupts in the cleft side rather than the noncleft side.

It seems that relationship between cleft and supernumerary teeth is not clear ,and etiologic factor of clefts may be attributed to have adverse effects on development of teeth buds and cause dental abnormalities. surgical interventions also can be assumed as a probable cause of dental abnormalities .the exact mechanism is not clear.

Presence of extensive defect in the upper face compared to lower face was so interesting, it can be because of that amniotic rupture sequence is more likely to affect the skull including the upper face rather than the lower face, because the forehead of the embryo is facing toward the amnion whereas the lower face is protected by upper face and body. the posterior alveolar area in the mouth is therefore less likely to be involved with amniotic bands.

we can only suggest that amniotic bands can cause disturbances in facial and dental developments.

Because of small number of similar reports and lack of some document of this patient further investigations therefore are required, we can only suggest that amniotic bands can cause dental and facial disturbances.

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