A Case of Amniotic Band Syndrome with Cleft Lip and Palate

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Abstract The patient was a 5-day-old boy born at 37 weeks and 4 days. At birth, an amniotic band encircling and constricting his left middle finger was noted, in addition to multiple anomalies including a right-side cleft lip and palate, a club foot, and syndactyly on the left hand. We performed cheiloplasty at 5 months, and palatoplasty at 18 months.

Keywords Amniotic band syndrome · Cleft lip and palate · Hotz plate · Club foot · Syndactyly

Introduction

Amniotic band syndrome is a rare congenital condition in which it is hypothesized that a fibrous band of amniotic tissue results in variable soft tissue derangement. This syndrome is a sporadic condition and is reported to occur in approximately 1 in 1,200–15,000 births [1–5]. The patient may present with craniofacial, thoracic, abdominal, and/or limb involvement. Here we report a case of amniotic band syndrome with cleft lip and palate.

Case Report

The patient was a 5-day-old boy who was born at 37 weeks and 4 days uneventfully (body weight: 2,375 g) on

September 8, 2005. The Apgar scores were 9 at 1 min and 10 at 5 min. At birth, an amniotic band encircling and constricting his left middle finger was noted, in addition to multiple anomalies including a right-side cleft lip and palate (Fig. 1), syndactyly on the left hand (Figs. 2, 3), and a club foot (Fig. 4). His mother is 43 years old and his father 38. No hereditary anomalies, including cleft lip and palate, were observed in his family. His mother did not stop smoking or drinking alcoholic beverages during the pregnancy. The patient used a Hotz plate to assist with milk feeding as well as to improve development of the maxilla. In the newborn period, he was neurologically intact with an appropriate response to stimulation and sleep-and-waking cycle. At four months he manifested age appropriate development and ultrasounds of his heart and abdomen were normal. There were no abnormalities in his blood examination results, ECG, cardiac echogram, abdominal echogram or chromosomal examination. We subsequently performed cheiloplasty by the Cronin's triangular flap method on his right upper lip at five months and palatoplasty by a modified push back method (palatal mucosal flap and muco-periosteal flap for closing of the cleft) at 18 months. Furthermore, orthopedists removed the constriction ring on his left-hand at 10 months.

Discussion

Forty-nine cases (27 males and 22 females) of oro-facial anomalies associated with amniotic band syndrome were reported in the literature over the past 20 years [1–15]. These cases included 37 cases of facial cleft and 9 cases of cleft lip and palate. Although the precise mechanism for development of oro-facial anomalies associated with amniotic band syndrome is not fully understood, two main theories of the pathogenesis of amniotic band syndrome,

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Fig. 1 Front view of the face showing the right-side cleft lip

Fig. 2 An amniotic band had encircled and constricted his left middle finger

namely the extrinsic model [16] and intrinsic model [17] can be employed for explaining the development of the oro-facial anomalies associated with this syndrome. The intrinsic model suggests that the anomalies and fibrous bands have a common origin, caused by perturbation of the developing germinal disc in the early embryo. The bands represent either sheets of macerated epidermis or are the result of a circumscribed area of imperfect histogenesis leading to sloughing off necrotic tissue. The aberrant tissue bands represent merely residues of an abnormal development process and not the cause. On the other hand, the extrinsic model suggests that the birth defect is caused by the action of fibrous amniotic bands. At present, the extrinsic model is widely accepted, with the sequence of events involving rupture of the amnion, followed by loss of

Fig. 3 X-ray of the syndactyly on the left hand

Fig. 4 Club foot

amniotic fluid and extrusion of all or parts of the fetus into the chorionic cavity. While trapped there, the fetus is subjected to compression due to the limited space, and the fetus' limbs and other body parts become entangled in the shriveled, rolled remnants of amnion. The exact nature of the anomalies can be accounted for by the timing of the rupture [18]. However, the extrinsic model cannot adequately explain the involvement of cleft palate and other anomalies of internal organs in amniotic band syndrome. Further investigation including genetic or genomic analysis may be necessary to understand the pathogenesis of amniotic band syndrome.

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