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Abstract

This article presents two cases of amniotic band syndrome associated with orofacial deformities, The surgical repair of the facial clefts and difficulties in overall management in our environment are further discussed

Introduction

- 1. Amniotic band syndrome (ABS):amniotic bands malformations that entangle fetal parts during intrauterine life
- 2. Results in various birth defects and deformities that reduce patient's life
- 3. ABS that involves the orofacial region may present as cleft lip or palate, asymmetric micro-ophthalmia, nasal deformity and oblique facial cleft. These craniofacial anomalies in ABS are typically often bizarre and frequently nonembryological in location
- 4. The exact cause of ABS is still unknown. This lack of direct evidence has led to the proposal of two main aetiological theories: the extrinsic and the intrinsic theory
- 5. The extrinsic theory :the band of ruptured amnion causes an extrinsic compression to the fetus
- 6. The intrinsic theory: germ cell deficiencies result in the malformations of the affected parts

The case report

Case one:

- 1. 15 years old female
- 2. No family history of ABS
- 3. It was reported that the mother took some traditional medicine for an unknown intermittent ailment during pregnancy
- 4. Clinical findings: unilateral, left-sided, complete cleft of the upper lip with rotated incisors
- 5. R't hand: amputation of index and middle finger, the little and ring finger were fused at the end of finger.
- 6. L't hand: the index, middle and ring fingers terminated at interphalangeal joints
- 7. An amniotic band constriction at the right leg with lymphoedema
- 8. The hallux and the second and third toes of both feet were amputated at the distal ends, while the fourth and fifth toes were slightly developed
- 9. The lip defect was repaired by the Millard rotation advancement procedure





Case two

- 1. 9 years old female
- 2. prematurely at home by a traditional birth attendant
- 3. No family history
- 4. Clinical findings: a depressed scar, about 2 cm in diameter extending from the apex of the v-shaped cleft of the lower lip to the tragus of the right ear was found
- 5. Also a right-sided commissural cleft of about 3 cm in length
- 6. The Left side, an oblique facial cleft that extended from the lateral end of the upper lip to the left lower eyelid
- 7. There was a deformity of the lower lacrimal canaliculus
- 8. The anterior wall of the maxilla was hypoplastic
- 9. The right side cleft of the lower lip was repaired by the V-plasty and the

right-sided commissural cleft was repaired by the linear suturing technique

- 10. Six months later the patient was recalled for the repair of the oblique facial cleft
- 11. The lower-eyelid part of the cleft was repaired with Z-plasty, while the closure of the labiomaxillary cleft was achieved by adjoining soft tissues in layers







Discussion:

- 1. ABS is a known cause of fetal malformations, and the anomalies range from mild deformities to severe ones that may be incompatible with post-natal life
- 2. The exact incidence is unknown(early pregnancy?)
- 3. Compression from these bands will then result in deformities of the

- developing extremities, trunk and head
- 4. In these our case reports, the clinical presentations of digits and toes amputation, leg constricting band with its associated lymphoedema, facial band and clefts could be a result of local compression
- 5. Therefore, the present case reports may further support the concept of extrinsic theory in the aetiology of ABS
- 6. The severity of the band compression on the developing fetus could determine the morphology of the deformity. When bands are superficial, only skin indentation occurs, as it was noted in case 2 on the right facial region. Deeper circumferential bands may cause lymphatic obstruction leading to oedema distal to the band, as shown in case 1
- 7. Tighter constricting bands have also been noted to cause the narrowing of underlying bone or the amputation. The case 1 is a good example
- 8. Although severe craniofacial abnormalities caused by ABS are often incompatible with life, a number of these children may still survive even till school age
- 9. In the case 2, The oblique facial cleft, right-sided commissural and lower-lip clefts in one single p't is a rare and unique ,strongly supports the hypothesis that amniotic bands might have been the etiological factor, especially with the presence of lateral oro-ocular cleft
- 10. In the Case 1, the lip defect was successfully repaired by the Millards rotation advancement procedure, and the second case required an advanced facial reconstructive procedure

Conclusion

• The presented cases with peculiar features of orofacial clefts, reduction defects of limbs, leg constriction band with lymphoedema and rare lower-lip cleft support the ideas that these deformities are likely related to ABS, and help us to known and treated in the future

題號	題目	
1	本篇作者支持哪個 theory 造成 ABS?	
	(A) Extrinsic theory	
	(B) Intrinsic theory	
	(C) Outer theory	
	(D) Inner theory	
答案(A)	出處:本篇 journal	
題號	題目	
2	Case 1 的 Lip defect 是用哪種術式治療?	
	(A) Amputation	
	(B) Z-plasty	
	(C) V-plasty	
	(D) Millards rotation advanced procedure	
答案(D)	出處:本篇 journal	