



A New Approach to Amniotic Band Disease

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Abstract

Background/Aim: Amniotic band disease (ABD) is a rare condition, generally associated with three types of anomalies: malformations, deformities and amputations. Several theories have been put forward, the extrinsic theory being the most widely adopted. The aim of the study was to clarify the pathophysiology of the disease, to propose a new classification and to precise the therapeutic indication through a radio-clinical description of seventy-five amniotic bands in ten patients.

Methods: This was a descriptive radio-clinical and surgical study of 75 amniotic bands in amniotic disease. The clinical features were used to describe the type of amniotic band: deep (impeding vascular circulation, compressing soft tissues and bone) or superficial cutaneous with a good prognosis. The shape of the band was described as semicircular, circular, heliocoidal or complex figure of two linked circles shaping the number 8 flat/bended. There were 5 mild constrictions without lymphoedema, 28 moderate (constrictions without lymphoedema in 7 cases and severe constrictions with lymphoedema in 21 cases) and 41 amputations (one leg and 40 fingers and toes). Surgical treatment involved 16 deep constrictions and three feet.

Results: Resection of the amniotic bridges gave an acceptable cosmetic result, but lymphoedema persisted. Surgery on both feet enabled walking with plantigrade support. However, the Ponseti technique was used on a child with a severe and complex heliocoid constriction. He developed ischaemia, requiring immediate removal of the cast. Digital separation of the fingers restored a functional hand in particular the pauci-digital pincer, but the aesthetics were judged insufficient by the parents.

Conclusions: Based on presented findings, amniotic limb disease should be classified into three types: 'Type A' with a good prognosis, 'Type B' subdivided into six sub-types and "Type C" defined by circular or complex constriction. The classification provides a therapeutic orientation. Depending on the type of the constriction band of the disease, different strategies can be envisaged: therapeutic abstention, cosmetic surgery or emergency limb salvage surgery.

Key words: Amniotic band syndrome; Amputation, congenital; Extrinsic theory; Constriction, pathologic; Classification.

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Introduction

Amniotic band disease (ABD) is a rare condition, accounting for 1/15,000 cases. It was first described by Montgomery in 1832.¹ It is a poor-

ly understood congenital malformation, due to strangulation of the organs by an amniotic fibrous band. All parts of the body are affected,

including the skull, face, neck, trunk and musculoskeletal system. It generally associates three types of anomalies: amputations, deformities and malformations. Several theories have been put forward, the extrinsic theory being the most widely adopted.²

The aim of the study was to clarify the pathophysiology of the disease, to propose a new classification and to precise the therapeutic indication through a radio-clinical description of seventy-five amniotic bands in ten patients.

In article published in the journal *Foot and Ankle Surgery*,³ authors' opinion on the pathophysiology of the disease and even modified Weinzweig's (1994) classification was presented.⁴ In presented article, we would like to propose a new ra-

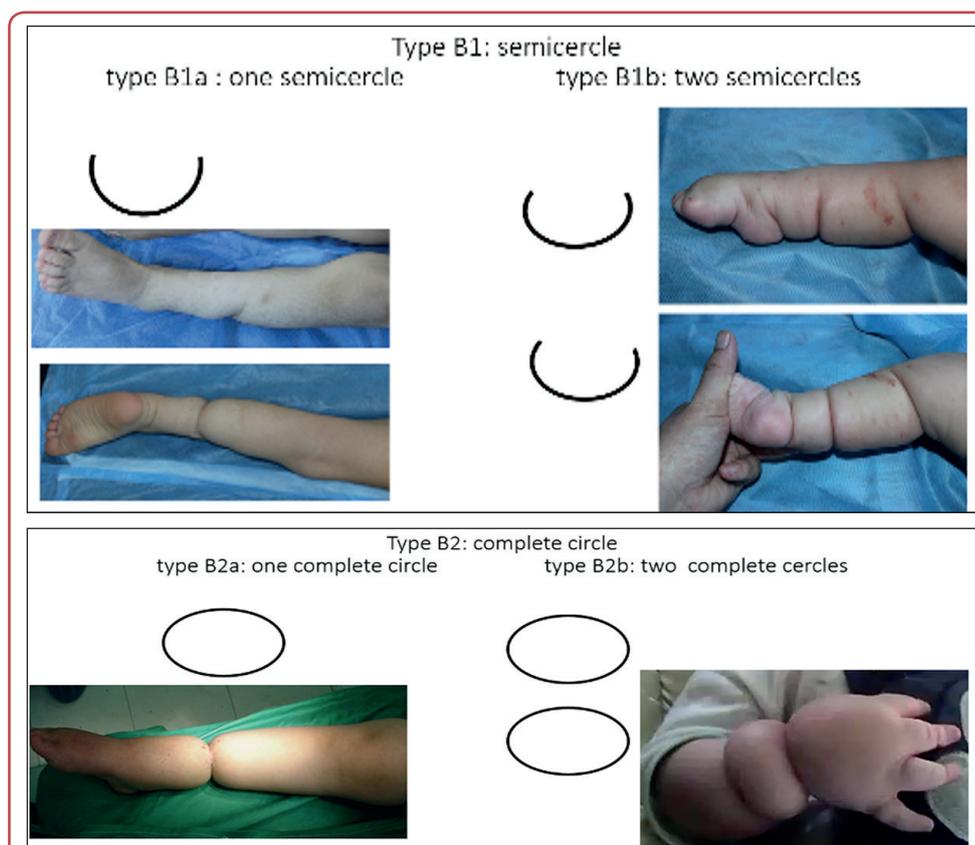
dio-clinical classification of the disease, based on data from the literature and the radio-clinical description of 75 amniotic bands in ten patients from our series, affecting different parts of the body: the upper limbs, the lower limbs and the face. It is also essential to review the pathophysiology of the malformation, particularly in the fingers, toes and legs.

Amputations, growth disorders and acrosyndactyly of fingers and toes have been observed. The mechanism of apoptosis in the genesis of fingers and toes is a factor to be taken into consideration when estimating the date of onset of constriction.⁵ The tools used are simple: visual observation of bands, photos, X-rays and intraoperative macroscopic findings.

Methods

This is a descriptive radio-clinical and surgical study of 75 amniotic bands in amniotic disease. Microsoft Excel was used for quantitative data analysis. The clinical features were used to describe the type of amniotic band: deep (impeding vascular circulation, compressing soft tissues

and bone) or superficial cutaneous with a good prognosis. The shape of the band was described as semicircular, circular, heliocoidal or complex figure of two linked circles shaping the number 8 flat/bended (Figure 1).



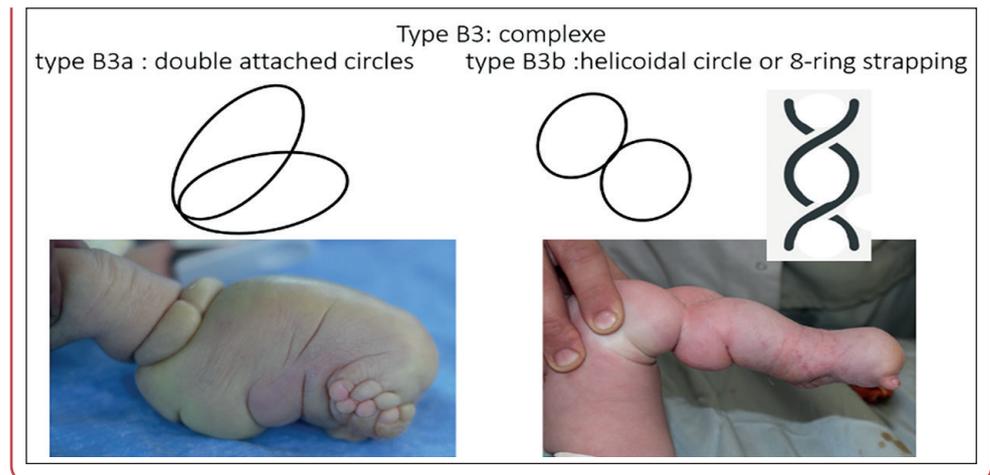


Figure 1: The three amniotic band subtypes: semicircular, circular and helical

There were 75 locations, ranging from amniotic bands constrictions to amputations. Nine children and one adult, six males and four females, aged between 10 days and 29 years. An average of 7.5 bands per patient. Involvement was multifocal, affecting the upper limbs, lower limbs and the face. The diagnosis was clinical, based

on the presence of at least one amniotic band, with or without amputation. Other congenital malformations, including limb agenesis and symbrachydactyly,⁶ were therefore eliminated. Weinzwieg's (1994) classification was used to divide the 75 locations (74 cases include bands affecting the limbs, 1 case includes the face).



Figure 2: Multiple localisations of amniotic bands in a 5-year-old child, localisation on his right hand shows a band taking the last 4 fingers. Surgery to release the fingers was performed with the postoperative result.



Figure 3: Child aged 10 days, operated on for pre-ischaemic voluminous foot, with open leg fracture, classified as a B3b foot type. Reviewed 6 years later with follow-up x-ray.

Table 1: Weinzwieg's (1994) classification used to divide the 75 locations

Weinzwieg's (1994) classification	75 locations (1 in the face)
Mild constrictions without lymphoedema	5
Moderate constriction	28
Amputation	41

The location of the bands in terms of the limbs concerns 74 constrictions; there were 5 mild constrictions without lymphoedema, 28 moderate (constrictions without lymphoedema in 7 cases and severe constrictions with lymphoedema in 21 cases) and 41 amputations (one leg and 40 fingers and toes) (Table 1).

Surgical treatment involved 16 deep constrictions (including fingers in 3 hands and 3 feet) (Figure 2 and 3). It should be noted that the 29-year-old woman with facial damage did not undergo surgery. The technique involved resection of the bands and skin closure with Z-plasty. The wounds concerned were deep circular bands with lymphoedema and distal deformity in one or two stages.

The thigh (once), the leg (three times), the wrist (twice) and fingers (nine glued fingers in two children and a single finger in another child) were found. It was technically difficult to separate the fingers in both children. Intraoperatively cartilaginous bone fusion was observed on two occasions and was not visible on standard preoperative radiographs.

Four feet were treated, two with *varus equinus* clubfoot were treated surgically and the third with a complex band, (Figure 1: type B3b), was treated using the Ponseti method. Finally, the fourth child had a preischemic neonatal foot, voluminous with severe constriction, skin opening and fractured leg bones (Figure 3). He underwent emergency surgery, which involved wound trimming, resection of the wound edges, release of the posterior tibial vascular axis, stabilisation of the tibia with an intramedullary pin and cosmetic repair of the foot.³

Results

There were thirty-one bands in the upper limbs. Twenty-nine bands were found in the fingers and two in the right wrist (Figure 4). The amniotic band was found seventeen times in the fingers (nine on the right side and eight on the left). Twelve digital amputations, (six on the right and six on the left) (Figure 4). A single thumb was affected by a semicircular dorsal band.

Forty-three lower-limb bands involved amniotic bands on the thighs (three cases) and eleven on both legs, with one mid-diaphyseal amputation of the left leg. Twenty-eight amputations (eight on the right and twenty on the left side) were

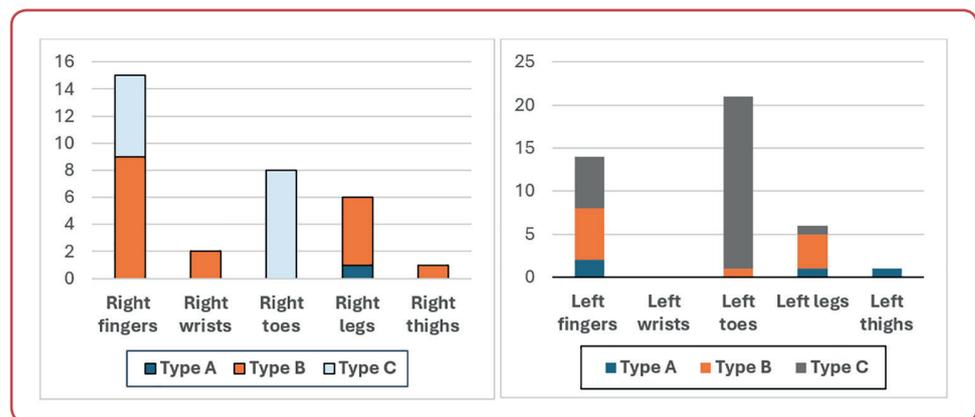


Figure 4: Localisations of amniotic bands

recorded on the toes (Figure 4). The face was affected only once with a particular amniotic naso-labio-palatal band, the 29-year-old woman's nose was flattened on the right side with a very visible skin furrow, the upper lip was the site of a depression and right hypotrophy. The cleft palate was complete and wide, taking up the entire length of the palate. Eating and speech were impaired.

Among the thirty-three amniotic bands found on both limbs, twenty-eight were known to be moderate hemicircular, without lymphoedema (7 occurrences), deep and severe circular impeding lymphatic or even blood circulation (21 occurrences) and five were superficial cutaneous, with no distal repercussions. Amputation was found forty-one times.

It was considered that it is important to distinguish between the shapes of deep semicircular and deep circular bands. Therefore, seven semicircular bands were identified, either unifocal (1 instance) or bifocal (6 instances). Nineteen times they were completely circular, unifocal (16 times) or bifocal (3 times). They sometimes took the shape of a figure eight folded in half (1 time) and sometimes a helical shape (1 time) (Figure 1).

Resection of the amniotic bridges gave an acceptable cosmetic result, but lymphoedema persisted. Surgery on both feet enabled walking with planigrade support. However, the Ponseti technique was used on a child with a severe and complex heliocoid constriction. He developed ischaemia, requiring immediate removal of the cast. Digital separation of the fingers restored a functional hand; in particular the pauci-digital pincer, but the aesthetics were judged insufficient by the parents.

Discussion

The literature review showed that publications concerning amniotic disease are rare. Few classifications have been used, such as Hall's 1982 classification⁷ into three types: mild constriction without lymphoedema, moderate constriction with lymphoedema and severe constriction with amputation.

Weinzweig's 1994 work is more detailed, with four types of increasing verity: mild, moderate and severe constriction with lymphatic, venous or even arterial compression and finally antenatal amputation.⁴ To our knowledge, no author has described or defined the type of superficial or deep amniotic band, nor its hemicircular, circular, complex figure similar to the number "8" or heliocoidal shape (Figure 3). This is because the correlation between the appearance of the amniotic band and the vascular and dysmorphic consequences downstream of the constriction are closely linked. For this reason, based on the 75 constrictions of patients and a review of the literature, authors identified and propose a new classification:

Type A: superficial skin constriction, with a good prognosis.

Type B: deep constriction of increasing severity, with or without lymphoedema. This category is subdivided into six sub-types:

Type B1: the constriction is semicircular, without lymphoedema, uni-focal 'Type B1a', bifocal 'Type B1b'.

Type B2: the constriction is circular, with lymphoedema, uni-focal 'Type B2a', bifocal 'Type B2b'.

Type B3: the constriction is complex and severe, with lymphoedema and pre-ischemic

state, with normal, deformed or fractured bone. The band taking the form of a figure-8 'Type B3a', or heliocoidal 'Type B3b'.

Type C: it is defined as neonatal amputation.

Superficial constriction is defined by skin involvement, but no involvement of the subcutaneous fat layer. Deep and severe constriction separates the subcutaneous fat layer, with the skin sticking to or coming into contact with the aponeurosis. It impairs lymphatic circulation, sometimes venous and in the final stage, arterial circulation. Bone damage, such as deformities or fractures, is a sign of severity.

In terms of the hand: acrosyndactyly is defined as distal fusion of the fingers with proximal fenestration, it is due to an arrest in the process of apoptosis during embryogenesis.⁵ This process begins between 6 and 8 weeks. Moreover, the programmed death of interdigital cells occurs from proximal to distal fingers. This is why the fingers are fused distally. We can therefore easily deduce the date of onset of the constriction of the fingers by the amniotic band, which occurs after this date, ie after 6 or 8 weeks. Questioning pregnant women about the period of natural fluid loss confirms this opinion. Several varieties of fenestration in presented patients were found: punctiform, linear and complete. This observation suggests that the date of the constriction is early in the punctiform and linear forms and late in the complete forms. Concerning the number of occurrences of the band constriction at the level of the lower limbs (43 times) is superior than the upper limbs (31 times); this is, perhaps, due to the distal proximity of the lower limbs compared to the upper ones which are separated by the trunk body. This finding highly agrees with data obtained from the literature.⁸ As for the rarity of the thumb involvement (1 time/74), this can be explained by its short size and above all by its high anatomical position in relation to the rest of the fingers.

In terms of the legs, it has been observed, in

two children, one was born with an amputation of the left leg and a posterior semicircular band of the right leg. The other child was born in the pre-ischaemic stage with an open fracture of the right leg and a hemicircular band of the left leg. Our thought concerning these two cases is that both legs were tied prenatally by a single band with severe constriction. This is what is called "The tied child by a single cord", the explanatory drawing clarifies this point of view well. Prenatal amputation following band constriction has been reported by several authors (Torpin and Faulkner)⁹ and reinforces the extrinsic theory of disease. Shipp¹⁰ reports a case of a newborn decapitated by an amniotic band, while Benirschke¹¹ describes the amputation of a newborn's foot, the macerated little foot having accompanied the placenta during delivery. The age of amputation was estimated at 20 weeks.

Concerning the face: the involvement of the skull and face has been mentioned in several articles, some of which consider it to be a malformation associated with amniotic disease, while others such as Kaplan¹² assert that it is part of the latter. We agree with this view and the case in presented series confirms this hypothesis. On the face of the woman, stigmata were found of an amniotic band tracing an oblique naso-labio-palatine line with a cleft palate.

To explain this, the first is necessary to look at the embryogenesis of the face and the date of appearance of the nose, mouth and palate. According to Carlson¹³ in his sixth book, the nose begins to develop at eleven weeks and the mouth (lips and palate) at the eighth week. He also states that intra-uterine thumb sucking and swallowing of amniotic fluid take place during this period. It therefore appears that the foetal face was constricted by an amniotic band with its mouth half-open during organogenesis of these organs, ie around the eighth and eleventh weeks.

The classification authors propose provides a therapeutic orientation. Depending on the stage

Table 2: The therapeutic indication according to the typology of the amniotic band

Types	A	B			C		
Sub-types		B1		B2		B3	
		B1a	B1b	B2a	B2b	B3a	B3b
Therapeutic indication	Abstention	Surgical treatment				Limb prosthetics	

of progression of the disease, we were able to envisage different strategies: therapeutic abstention, cosmetic surgery or emergency limb salvage surgery.

Therapeutic abstention in Type A. Z-plasty surgery is aesthetic in hemi-circles without lymphoedema, in Type B1. It is imperative in circular forms with lymphoedema in type B2 and urgent surgery in type B3. A prosthesis is required for type C (neonatal amputation) (Table 2).

Concerning the hand: finger release should be performed as soon as possible, or even as a matter of urgency, as adhesions interfere with digital growth and function. Several hypotheses are then possible concerning the consequences of strangulation, such as growth problems, lymphoedema or immediate amputation of the fingers.

About the feet: Ponseti's technique is not recommended in forms with lymphoedema, in types B2 and B3. There is a risk of worsening lymphoedema and limb ischaemia. Aouran¹⁴ demonstrated in his article that the presence of the ipsilateral band does not affect the efficacy of the Ponseti technique, but he did not take into consideration the type of the amniotic band. Deep or superficial, hemicircular, circular or complex and with or without lymphoedema. The case cited in presented series, developed ischaemia after plaster cast immobilisation, presented with a deep helical complex band. Hence the importance of classifying the type of amniotic band, sometimes necessitating treatment of the fibrous band before correction of the clubfoot on ABS. This makes it essential to propose a classification based on the above-mentioned parameters.

In his article, Demetri⁸ considers that equinus clubfoot is a congenital malformation associated with amniotic disease and that the ipsilateral presence of the amniotic band does not explain its occurrence. The opposite is true: one of the theories of the pathophysiology of idiopathic equinus varus foot is the theory of vascular ischaemia. And we believe that equinus clubfoot is the direct consequence of prenatal ischaemia, particularly of the posterior compartment. It should be noted that the 3-foot treated in presented series had distal toe amputations, which reinforces authors' opinion.

Conclusion

The literature review showed that publications concerning amniotic disease are rare. Few classifications have been used. Based on findings, authors propose classification of amniotic limb disease into three types: type A with a good prognosis, type B it is subdivided into six sub-types and type C it is defined by circular or complex constriction. Proposed classification provides a therapeutic orientation. Depending on the stage of progression of the disease, different strategies can be envisaged: therapeutic abstention, cosmetic surgery or emergency limb salvage surgery.

Ethics

This study was approved by Ethical Committee of Batna 2 University, Algeria, decision No 26/CE-DUB2/2025, dated 24 May 2025. Study was conducted in compliance with ethical principles and current regulations. All participants provided informed consent before inclusion in the study. All data were processed anonymously and confidentially.

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Conflicts of interest

The authors declare that there is no conflict of interest.

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Data access

The data that support the findings of this study are available from the corresponding author upon reasonable individual request.

Author contributions

Conceptualisation: NK
Methodology: NK, HA
Data curation: NK, HA
Writing - original draft: NK
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