AMNIOTIC BAND SYNDROME OF THE TRUNK: A CASE REPORT

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Keywords Amniotic band syndrome, skin, child.

Abbreviation **ABS** = amniotic band syndrome.

Case report. A 3-year-old female, one of monozygotic twins, presented with a depressed, circumferential, and mildly hypopigmented band around the trunk, which had been present since birth (Figs. 1, 2). The linear skin crease was located below the umbilicus anteriorly, while appearing slightly higher on the back. The lesion was more pronounced when the patient was standing compared to the supine position. No erythema or induration was observed. The patient was completely asymptomatic. The medical history provided by the mother revealed no functional impairment, no history of atopy, and no relevant family history. The patient's growth and development were within normal limits. Apart from an infantile hemangioma on the right flank, the remainder of the physical examination was unremarkable. The clinical and anamnestic data led to the diagnosis of amniotic band syndrome (ABS).

Ultrasonography showed discrete, focal skin thickening beneath the lesion, along with a minimal localized septum in the subcutaneous tissue. The underlying muscular fascia appeared slightly prominent (Fig. 3). No additional abnormalities were identified. These findings were consistent with ABS, although literature regarding the radiological findings for this condition remains limited.

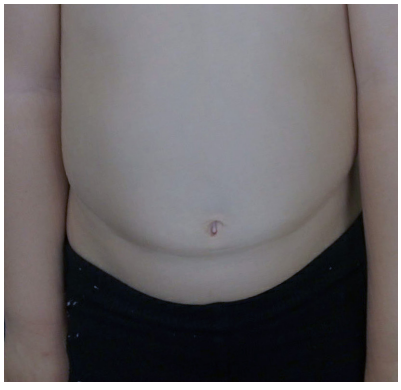

Fig. 1

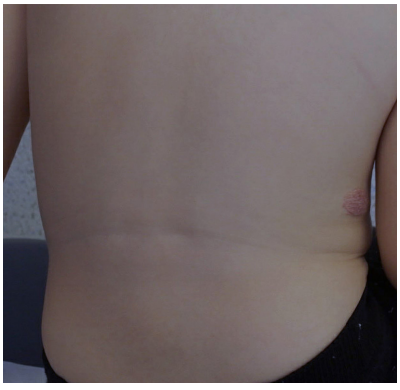

Fig. 2

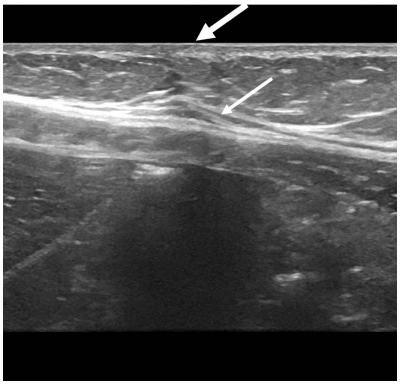

Fig. 3

Figs. 1, 2, 3: Circumferential amniotic band syndrome of the trunk (Figs. 1, 2). Ultrasonography (Fig. 3) shows focal skin thickening and a minimal localized subcutaneous septum.

Discussion. Amniotic band syndrome (ABS) is a condition with a broad spectrum of phenotypic presentations. The most common manifestations of ABS include constriction rings, limb or digital amputations, and acrosyndactyly (1, 2). Less frequent presentations include craniofacial anomalies, such as encephalocele and facial clefts, as well as thoracic and abdominal wall defects, including ectopia cordis and intestinal extrusion. Ocular involvement, such as hypertelorism, ptosis, and corneal

opacities, is a common feature of the disease (1). Potential associated anomalies include lymphedema, clubfoot, limb-length discrepancy, and phalangeal hypoplasia (2).

Abdominal involvement in ABS is rare, with only a limited number of cases described. These lesions may present circumferentially (3, 4), but can also manifest differently (5). The constriction band appears as a groove located either above or below the umbilicus (3-6). The depth of the lesion may vary, but it never extends beyond the first fascial layer (3). In circumferential cases, the portion on the back is slightly higher than the anterior aspect, as observed in our patient (3, 4). Consistent with previous cases, no functional impairment was reported by our patient (3, 4). Notably, both our case and previous reports indicate that the affected individual is one of a pair of monozygotic twins, in whom the other twin does not present with the condition (4, 6).

The pathogenesis of ABS remains incompletely understood. Two main theories have been proposed (1). The extrinsic theory, or amniotic band theory, was first described by Torpin in 1965 (5, 7). According to this hypothesis, these lesions are caused by an early rupture of the amnion, which leads to the loss of amniotic fluid and the formation of fibrous strands that entrap fetal parts between the amnion and chorion. This can result in vascular compression and tissue necrosis (1, 7). Amniotic bands consist of inelastic fibrous strands, which allows them to exert a ligation-like constrictive effect (3, 5). The underlying causes of the initial amniotic rupture remain unclear (1). Of note, this model cannot account for all possible manifestations of ABS, such as syndactyly or facial clefts (7).

Conversely, the intrinsic theory, or developmental defect theory, proposed by Streeter in 1930, suggests that the lesions stem from an embryonic genetic defect. According to this hypothesis, the observed anomalies are caused by disruptions in embryonic development rather than a secondary mechanical process (5, 7, 8). To date, no genetic predisposition has been identified (8).

The diagnosis of ABS can be established either prenatally or postnatally (1). Prenatal diagnosis in the first trimester remains challenging. In the second and third trimesters, however, major abnormalities can be identified more easily via ultrasonography (2). A fixed fetal position, persisting despite maternal movements during the ultrasound examination, may suggest fetal entrapment by amniotic bands. When visible, amniotic bands appear on ultrasonography as thin, linear hyperechoic structures, although they are only rarely visualized. In complex cases or when intervention is being considered, fetal magnetic resonance imaging (MRI) can serve as a useful adjunctive diagnostic tool (1).

In the presence of ABS, the neonate must be thoroughly examined, with particular attention paid to the umbilical cord, digits, limbs, and the skin in general (1). Further investigations may include MRI and histological evaluation. In cases of trunk ABS, MRI typically reveals an intact abdominal wall with normal underlying visceral structures (8). Histologically, the lesions are characterized by a normal epidermis overlying a thickened dermis, with an increased percentage of hyalinized connective tissue (4, 8).

Because clear guidelines for the treatment of ABS are lacking, management must be individualized. A comprehensive physical examination is recommended in all cases. When indicated, additional radiological evaluation and consultation with a surgical team may be considered (1). Surgical treatment is not essential for superficial constriction bands that do not entail functional consequences (2). In cases where vascular supply is compromised, surgery may become necessary (1, 2). Surgical intervention involves the excision of the circumferential lesion under general anesthesia, with a 1-2 mm margin of intact skin. Z-plasty or W-plasty techniques are utilized to prevent secondary scar contracture (8). In cases with a subcutaneous tissue deficit, rectangular plasty and dermo-hypodermal rotation flaps can be employed. This operative treatment can be performed starting from 3 months of age; delayed excision has been associated with slower wound healing and a higher number of complications, such as secondary eczema at the lesion site (2).

However, Bahadoran et al. suggest managing trunk ABS cases via follow-up until growth slows down, and that surgical intervention may be reserved solely for patients with functional impairment (4).

Conclusion. Amniotic band syndrome of the trunk is an exceedingly rare condition that requires a thorough clinical examination to rule out other malformations and, occasionally, corrective interventions.

Conflicts of interest

The authors declare that they have no conflicts of interest.

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