

Congenital Constriction of Leg Causing Gangrene in Newborn: A Rare Case Report

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ABSTRACT

A newborn baby with a gangrenous limb has been rarely reported in literature. It has been said to be due to constricting amniotic band sequence. We hereby report such a case born to 36 weeks primiparous female who was not booked antenatally. There was a constriction band visible in the middle of leg. There were no other associated anomalies. The limb was subsequently amputated and baby survived. It reflects the primitive health seeking behaviours in our country.

Key words: Newborn, Gangrenous Limb, Amniotic Band Sequence.

INTRODUCTION

Congenital constriction of leg is a well-known condition in orthopaedics but a newborn with gangrenous limb is a rare finding for an orthopaedic surgeon. We hereby report such a case.

CASE REPORT

A 36 weeks pregnant lady came to our institution in labour. She was not booked previously in ante natal department and had no ultrasonography report with her. She delivered subsequently within 2 hours of coming to institution. It was a breech presentation and there was no difficulty in labour. The medical team was surprised to see the swollen black leg of baby. APGAR score at one, three and five minutes was normal.

On examination baby weight was 2.5 kg. A band of constriction was visible in middle of left leg. Foot and leg below the constriction were blackish and swollen. (Fig 1)

It appeared as if baby was wearing black leather boot. All other limbs were normal and there was no other congenital anomaly. Orthopaedic surgeon advised amputation but attendants did not consent immediately.


Baby was shifted to nursery. On blood investigations baby's total leucocyte counts were raised (16,800). Baby's blood group was AB -ve and mother's was AB+ve. Mother was non diabetic. There was no other abnormality in blood investigations. X-Ray of the affected part was done and there was fracture of both bone leg just distal to the constriction. The limb was amputated subsequently on day four after informed consent of parents. Further course was uneventful and baby was discharged on day 12 after wound healed.

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Article History:

Received: 07-01-2016, Revised: 18-01-2016, Accepted: 04-03-2017

Access this article online	
Website: www.ijmrp.com	Quick Response code 
DOI: 10.21276/ijmrp.2017.3.2.054	

DISCUSSION

Amniotic band syndrome, also called amniotic band sequence (ABS) refers to spectrum of congenital anomalies including disruptions, deformations and malformations, in which mostly limbs are affected. It occurs in 1 in 1200 to 1 in 15000 live births.¹

There are two main theories for the pathogenesis of ABS, and are referred to as the "extrinsic model" and the "intrinsic model." The intrinsic model was proposed by Streeter in the 1930² and suggests that the anomalies and the fibrous bands have a common origin, caused by a perturbation of developing germinal disc of the early embryo. Later, the Torpin's model of the 1965,¹ the "extrinsic theory", suggested that the birth defects are caused by the action of the fibrous amniotic bands with the sequence rupture of the amnion, followed by loss of amniotic fluid and extrusion of all or parts of the fetus into the chorionic cavity. The fetus' limbs, while trapped are subjected to vascular compression and then necrosis. The condition can be diagnosed on antenatal ultrasound and in utero techniques and salvage of limbs have been reported.^{2,3}

The index case had many unique features. Firstly, presentation of newborn with gangrene of the limb at birth and that to with fracture at that level has never been reported in India. The typical swollen leather boot appearance and fracture at the same level indicate extrinsic compression after fetal parts were already developed.

This case brings to our attention the failure of our health care services to outreach people. Despite having tertiary care health services in our country, health seeking behavior's are still primitive in some parts of our country due to illiteracy and poverty.



**Fig 1: Band of constriction was visible in middle of left leg.
Foot and leg below the constriction were blackish and swollen.**

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Source of Support: Nil.

Conflict of Interest: None Declared.

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Cite this article as: Hemant Jain, Ashok Singhvi, Abhinav Bhardwaj, Kishore Raichandani. Congenital Constriction of Leg Causing Gangrene in Newborn: A Rare Case Report. Int J Med Res Prof. 2017; 3(2):266-67. DOI:10.21276/ijmrp.2017.3.2.054