

## *Congenital Ring-like Constriction due to Annular Bands: Report of Two Cases*

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### Summary

**Onuba O and Efem S. Congenital Ring-like Constriction due to Annular Bands: Report of Two Cases.** *Nigerian Journal of Paediatrics* 1985; 12:135. Two cases of congenital ring-like constriction due to annular bands are reported. One was a 3-month old Nigerian infant who was treated surgically and the other, a 7-day old caucasian who was managed conservatively. Congenital annular bands and amputations are uncommon forms of deformity whose aetiology was previously thought to be related to genetic and dietary factors. Recent evidence from animal experiments and human placental studies, however, seem to implicate amniotic rupture as the main cause.

### Introduction

CONGENITAL annular bands are recognised causes of ring-like constriction around limbs.<sup>1</sup> In severe cases, such constrictions can lead to limb amputations.<sup>2 3</sup> Other anomalies like syndactyly, club foot, craniofacial defects, (cleft lip, cleft palate, cleft nostril) and congenital pits have also been reportedly caused by these bands.<sup>4 6</sup> The origin of these bands is uncertain. Single gene inheritance defect,<sup>5</sup> dietary deficiency<sup>4 7</sup> and loss of amniotic fluid<sup>8</sup> have been incriminated. Lesions produced by these bands are very rare. The incidence among livebirths varies between 1 in 10,000 and 1 in 15,000. Experience with the problems created by the

bands is therefore, uncommon. It is in realization of this that we report two cases of limb constriction due to annular bands seen recently at the University of Calabar Teaching Hospital (UCTH), Calabar.

### Case Reports

#### Case 1

BE was a 3-month old boy when he was referred to the UCTH from a peripheral hospital. He was a product of full-term pregnancy. The mother had minimal antenatal care in a local maternity home and there were no prenatal problems such as bleeding or liquor drainage. She could not recollect taking any traditional herbs or medication during the pregnancy except conventional anti-malarial drugs. Delivery at home was uneventful. The mother was 21 years old and the father, 32 years old. There was no consanguinity. This case was the first issue of the union.

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At birth, the baby was a normal looking child except for the cord of white tissue extending from the left big toe outwards and upwards and forming a tight constriction ring around the left calf just above the ankle (Fig 1). The nail of the big toe was absent and the four lateral toes showed significant webbing. The foot was also mildly clubbed (Fig 2). The segment of the leg below the constriction had normal colour and sensation and was as warm as the portion above. Power rating of the muscles showed only slight reduction in power in this lower segment. A 2-staged z-plasty, involving lifting multiple myocutaneous flaps while at the same time excising the constriction tissue, was performed with primary healing and cosmetic success.

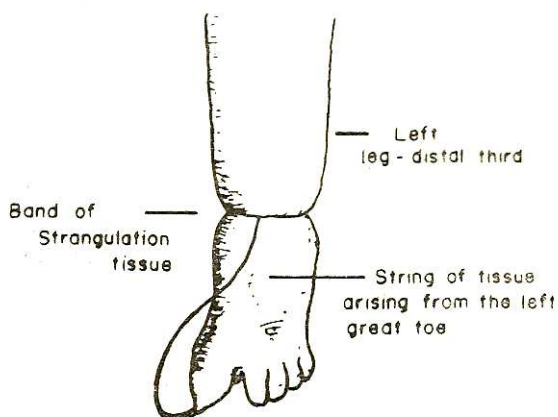


Fig 1. An artist's impression of the original band of tissue from the left big toe encircling the lower part of the left leg.

#### Case 2

The second case AB, was 7 days old when he was referred to UCTH. He was delivered at full term after an uneventful pregnancy. Antenatal care and delivery were at a private maternity hospital. Both parents were Europeans, the mother was aged 25 years and the father, 27 years. This was their first child. Family history of congenital annular band was absent. On examination, the baby looked normal except that there were constriction rings around the 2nd, 3rd and 4th toes of his left foot at the level of the proximal interphalangeal joints. The distal

parts of the toes were bluish and oedematous. The left foot was kept warm under the blankets for 48 hours after which the swelling of the toes completely disappeared and the colour became pink.



Fig 2. The constriction ring around the lower part of the left leg.

#### Discussion

The significance of annular bands as a cause of congenital ring constriction and congenital amputation was recognised as early as the beginning of the nineteenth century.<sup>10 11</sup> But the origins of these bands were not recognized. While Streeter<sup>12</sup> believed that the deformities caused by these bands were just a dysplasia due to imperfect embryogenesis, Barsky<sup>5</sup> postulated a single gene defect. Warkany<sup>4</sup> and Duraiswami,<sup>7</sup> based on their observations of experimental animals, held dietary deficiency especially of riboflavin and nicotinamide responsible for the bands. The role of dietary deficiency remains

unsustained since annular bands are as rare in the technologically developed countries where meticulous care is taken about diet during pregnancy as in the under-developed world battling with acute food shortage. The amnio-chorionic origin of these bands postulated by Poswillow<sup>13</sup> has been confirmed by others.<sup>6 8</sup> Kino<sup>8</sup> successfully reproduced constriction bands, congenital amputations and cleft palate in embryo rats by amniotic sac puncture on the 15th day of gestation. He observed haemorrhages from the marginal blood sinuses of the feet and attributed these to excessive contraction of the uterus following amniocentesis. Field<sup>14</sup> studied the microscopic details of placentae of 2 infants born with congenital constriction rings. The absence of the amnion in one case and the preservation of a broad structural remnant rolled up on itself in another case provided ample evidence of amniotic origin of the bands. One of the mothers had noticed a watery vaginal discharge at 4 months of pregnancy, although this was not observed in any of our cases.

No significant relationship has been demonstrated between the anomaly and drug ingestion, maternal viral illnesses in the first trimester, oligohydramnios or polyhydramnios. Moses<sup>1</sup> found a high incidence among first pregnancies, an experience that tallies with our finding. However, others have found it to be commoner among younger children in large families.<sup>2</sup>

A staged z-plasty operation aimed at restoring full function and to prevent future complications such as vascular insufficiency and elephantiasis-like limb was successfully performed in the first case. There is also room for conservative management as was pursued in the second case. Amputation should be avoided except in cases of gangrene.

Reports from Singapore,<sup>2</sup> a developing country like Nigeria, have shown that women regard congenital limb deformities in their newborn babies as a visitation from the gods especially when these deformities are associated with intrauterine amputation. Experience with

isolated cases as in this report and from different communities, will aid diagnosis, correct misconceptions and foster good patient care.

#### Acknowledgements

We are grateful to Mrs Goldberg, the Librarian at King George Hospital, Ilford, England, for furnishing us with the reference material; our gratitude also goes to the Audio-visual unit of the University of Calabar for the photographs and diagrams and Prof CE Effiong for reading the scripts.

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Accepted 16 May 1985