Congenital Band about the Pelvis (A Case Report)

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Abstract

Congenital band is a rare collection of fetal malformations associated with fibrous bands that appear to entangle or entrap various fetal parts in the uterus leading to deformation, malformation, or disruption. Heredity, sex, and race seem to play insignificant role, and its etiologic explanation is still confusing and conjectural.

Congenital band about the pelvis is very rare and only five such documented cases are reported in the world medical literature. Now we present the sixth case of congenital band about the pelvis, treated with multiple Z-plasties, with particular consideration on the etiologic factors.

Key words: Congenital band, Fetal malformation

INTRODUCTION

Congenital band is a collective terminology of rare fetal malformations associated with fibrous bands that appear to entangle or entrap various parts in utero leading to deformation, malformation, or disruption. This anomaly is given many other names such as congenital constriction ring, annular band, and amniotic band syndrome, and has variety of clinical presentations.

Congenital band about the pelvis is an extremely rare entity, and only five such cases are reported in the world medical literature. A few interesting points could be extracted from the five cases. There was no sexual or racial inclination in those five cases. Three had the congenital bands above the pelvic brim, two below the pelvic brim. However, in all five cases, the bands appear to be higher posteriorly, likely following a specific somite.

None of these reports commented on the associated congenital anomalies in two cases. Schneider’s case had pilonidal sinus and cleft of soft palate, Izumi and Arnold’s had bands around two toes and club foot. Casaubon’s case had no associated congenital anomaly.

Only Evans’ case had family history that patient’s mother had a band around one toe.

Surgical correction was performed in three cases, all by Z-plasty. Two of these treated patients got pregnant and gave birth to sons without any congenital abnormality.

CASE REPORT

A 17-year-old Korean girl visited our clinic for a band-like depression around the waist (Figure 2) and short first, second, and third toes of the right foot. (Figure 1) She was born in 1969 as the younger of two siblings with no family history of congenital anomaly. The pregnancy and delivery were uneventful.

Physical examination showed depressed, constrictive, fibrous band all around the lower abdomen and back, and distal amputation of first, second, and third toes of the right foot, which proved to be absence of the distal phalanges by X-ray evaluation. Otherwise there was no remarkable abnormal finding.

In 1983, multiple Z-plasties after excising the band were performed for the release of constriction ring about the pelvis (Figure 2). The biopsy specimen showed the epidermis to be normal. The dermis was thick, composed of collagen bundles and elastic fibers, nearly normal.
ing the subcutaneous tissue.

The adnexal structures were normal. (Figure 5, 6) There was no more constriction after Z-plas-

DISCUSSION

Of five reported cases of congenital band ab-

out the pelvis, Izumi and Arnold’s case is similar
to ours. However, generalization of all six cases
(including our case) seems to be difficult; Sex,
race, and heredity appear to play a very limited
role. The congenital band about the pelvis does
not seem to offer a real functional constriction,
rather it seems to be a cosmetic problem be-

cause it allowed normal pregnancies with or

without Z-plasty in two cases. The location of
the bands is almost uniform, along a specific so-

mite, higher posteriorly. Four of six cases were

formed by Z-plasty. Since Stevenson’s sugges-
tion in 1946, Z-plasty has been the most com-

mon method for the release of congenital band.

There are extreme controversies on the etology
of congenital band. In 1930, Streeter propo-
sed his theory of defective germ plasm. After

studying 16 preserved laboratory specimens of

amniotic band syndrome he concluded that con-

genital band were due to developmental defects
of the germ disk and imperfect histogenesis in
the developing tissue lead to a localized scli-

rosis. Schneider supported Streeter’s view with

his microscopic findings of his case, which re-

semble the discission of Streeter of “the these...strands...proved to be the hyalinized pro-
duct of defective tissue.” Keith suggested that

Streeter’s fetal dysplasia was due to early placental circulatory disturbance. However,

Streeter’s view of intrinsic etiology failed to ex-

plain the inconsistencies of asymmetry, sporadic occurrence, and the absence of related internal anomalies.

An alternative theory for the etiology of con-

genital band was published in 1965 by Torpin.

After the study of the placenta of the patient

with congenital band he concluded mesodermal

band (amniotic and chorionic fibrous strings)
formed following rupture of the amnion could

encircle fetal parts and similarly produce the

characteristic constriction ring. In 1981, Mayou

and Fenton reported in their study of six cases

of facial cleft that an amniotic string was found

lying deep in the bilateral oblique facial cleft. In

1982, Opgrande suggested their case of con-
genital band to be the result of extrinsic pres-

sure produced by the fibrous band that was thought to be amnion on the microscopic ex-
amination. In 1975, Kino concluded in his study

using rats that the constriction ring was the re-
sult of excessive contraction of the uterine mus-
cle and hemorrhage from the marginal blood sinuses, and suggested the malformations were

not hereditary, but are probably produced by pre-
natal environmental factors. Although the majority of recent papers support

Torpin’s view that congenital band results from

extraamniotic, intrachorionic entanglement with

mesodermic strands, and that theory seems best

able to explain the broad spectrum of clinical fe-

atures, the etiologic explanations remain confus-
ing and conjectural, and need to be further clar-

ified.

We think a rather specific location of congeni-
tal band about the pelvis favors the Streeter’s

view of intrinsic etiology.

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체간에 발생한 선천성 수축반혼환

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선천성 수축반혼환은 서유성 링(band)가 자궁내 태아신체의 일부를 엎들지 않아서 변형 또는 절단을 유발하는 태아기형의 일종으로 이에 전후 annular band, congenital constriction band syndrome, congenital ring constriction, amniotic band syndrome, amniogenic band 등의 여러 명명으로 불리지 했다.

임상적으로는 많은 경우 다른 여러가지 기형을 동반할 수 있으며, 주로 사지에 발생하고, 두안 끈기형이나 대장 기형도 흔히 동반하는 것으로 알려졌으며, 여분 체부에 생기는 수축반혼환은 주로 홀리하여 현재까지 문헌상으로는 전세계적으로 5례만이 보고되었을 뿐이다.

이 자리는 요부 주위에 형성된 홀리한 체간 선천성 수축반혼환을 규모 데 Z-형성술로 치료하고 좋은 결과를 얻었기에 문헌고찰과 함께 보고하는 바이다.

LEGEND FOR FIGURES

Fig. 1. Distal amputation of right first, secnd, and third toes.
Fig. 2. Congenital band about the pelvis. Note the band is higher posteriorly.
Fig. 3. Postoperative view after multiple Z-plasties (abdomen).
Fig. 4. Postoperative view after multiple Z-plasties (back).
Fig. 5, 6. Thick dermis with collagen bundles extending below eccrine glands.