Congenital Clubfoot: Is It the Result of Compression or Moulding in Utero ?

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Abstract : *Two fetuses with obstructive uropathy had clubfoot diagnosed on ultrasound before 18 weeks of gestation in the presence of normal amniotic fluid volume. Both had normal karyotypes. Oligohydramnios in the presence of obstructive uropathy may merely be associated with, rather than be the cause of clubfoot. Therefore, congenital clubfoot is not always the result of compression or moulding in utero. (Thai J Obstet Gynaecol 1989; 1 : 139-42.)*

Key words : congenital clubfoot, compression, moulding

Clubfoot is a common birth defect with an overall prevalence of 1.2-3 per 1000 live births[1, 2]. Since Parker and Shattock[3] in 1884 and Browne[4] in 1934 made the observation that clubfoot was associated with oligohydramnios and, therefore, caused by intrauterine moulding, many papers have been published supporting this theory[5-8].

Others, however, have suggested the contrary that a regional growth disturbance is the cause of clubfoot[9-11]. The observation of such an abnormality in the absence of oligohydramnios would argue against the importance of amniotic fluid and the compression or moulding in its aetiology.

The author reports bilateral talipes equinovarus in 2 fetuses with obstructive uropathy but normal amniotic fluid volume in whom the limb defect was diagnosed sonographically before the development of oligohydramnios.

Materials and Methods

Two patients with obstructive uropathy were referred between 16-18 weeks gestation. Scans were performed using an Acuson 128 (Acuson, Mountain View, California), with a 5.0 MHz transducer. In both fetuses, ultrasound guided fetal blood sampling was performed for karyotype determination and

urine aspirated from the dilated bladder for measurment of electrolytes.

Amniotic fluid volume was termed normal if at least one pocket of amniotic fluid measuring 3 cm in its vertical diameter was identified[12].

Results

Obstructive uropathy was confirmed in both fetuses by ultrasound signs of dilated bladder, bilateral hydronephrosis and in one of them, dilatation of the urethra (Table 1). Clubfoot was diagnosed in both cases (Fig. 1). The volume of amniotic fluid was normal in all. Karyotypes were normal in both fetuses. Urine electrolytes were suggestive of some residual renal function[13].

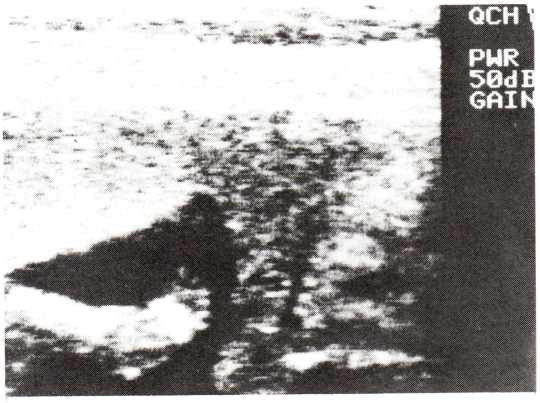

Fig. 1 Ultrasound showing clubfoot at 18 weeks gestation with normal amniotic fluid volume

One patient elected to undergo termination, while the other pregnancy ended in spontaneous abortion. Postmortem findings confirmed obstructive uropathy and bilateral clubfeet in both fetuses (Fig. 2).

Table 1 Clinical, ultrasound and pathology correlations

AGE	PARITY	GA (wk)	ULTRASOUND FINDINGS	AMNIOTIC FLUID VOLUME	KARYOTYPE	OUTCOME	PATHOLOGY
38	G1P0	18	Distended bladder Massive dilatation of penis, Megalourethra, *Bilateral clubfeet*	*Normal*	46, XY	Termination of pregnancy	Prune Belly syndrome, *Bilateral talipes*
27	G1P0	16	Huge dilated bladder, Bilateral hydronehrosis, *Bilateral clubfeet*	*Normal* Oligohydramnios (1 week later)	46,XY	Spontaneous abortion	Cystic dilatation of kidneys, Dilated ureters and urethra, *Bilateral talipes*

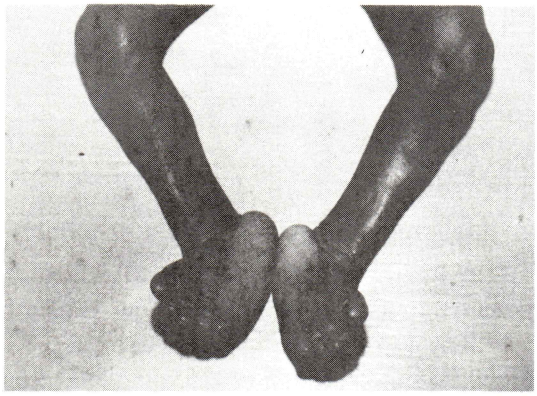

Fig. 2 Stillborn with bilateral clubfeet at 23 weeks gestation

Discussion

Both of the fetuses reported here had normal karyotypes. Reported incidence of abnormal karyotypes in fetuses with clubfoot ranges from 22-25 per cent[14, 15]. The sonographic detection of potential abnormality of clubfoot is clearly an indication for rapid karyotyping. In a series of 18 cases of talipes diagnosed by ultrasound, all of whom had normal ammiotic fluid, 83 per cent were associated with other abnormalities[14].

In these 2 cases, prenatal diagnosis of clubfoot was made in association with obstructive uropathy in the absence of oligohydramnios. One of the patients developed oligohydramnios the following week. However, they were both at or before 18 weeks gestation, when the contribution of fetal micturition to amniotic fluid is minimal. It has always been assumed that clubfoot, when diagnosed in the presence of oligohydramnios is due to the reduced volume of the amniotic fluid within the uterine cavity, holding the limbs in a fixed position[2-8]. This theory is based both on the association of clubfoot with intrauterine mechanical factors[8, 16, 17], and on the experimental creation of limb anomalies in animal models[18, 19]. The relevance of these latter studies to human idiopathic clubfoot is uncertain. No evidence exists, that mothers of children with clubfoot are selectively exposed to certain drugs or environmental conditions.

On the other hand, Dietz[9] has suggested, based on clinical findings, that the leg and the foot are invariably small in clubfoot, and a regional growth disturbance may be the cause of clubfoot. The more severe the deformity, the greater the reduction in foot and leg size. Even after adequate correction, deformity may recur during the major growing period of the foot. It is believed that clubfoot resulted from delay in the growth of tissues of the posteromedial compared to the anterolateral foot and leg. Other investigators have found a disproportionate amount of type 1 muscle fibres in many posterior and medial muscle groups and in several peroneal muscles[10, 11]. This suggestion that a regional neural abnormality may be present, since muscle fibre type is neurally determined.

Oligohydramnios was clearly not a causative factor in the two cases of clubfoot described here, which were associated with obstructive uropathy. Congenital clubfoot, therefore, is not always the result of compression or moulding in utero. Whether there are neuronal or tropic mechanisms common to both urinary tract and lower limb

abnormalities or not requires further investigation.

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