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Constriction band of the arm following insertion of a pleuro-amniotic shunt

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ABSTRACT

In a 23-week fetus with a large left-sided pleural effusion a double pigtail pleuro-amniotic shunt was inserted uneventfully. A healthy infant was delivered at 38 weeks' gestation. One end of the shunt was in the thoracic cavity but the other end was found wrapped around the upper arm resulting in a constriction band. There was no impairment of the vascular supply to the limb or in its motor or sensory function and by 6 months of age there was only a faint ring still apparent around the upper arm.

CASE REPORT

The mother was a healthy 22-year-old Caucasian who had previously had a healthy baby delivered by Cesarean section for cephalopelvic disproportion. In this pregnancy she was initially seen at 13 weeks' gestation for a first trimester scan. Although no structural abnormalities were detected, the fetal nuchal translucency thickness was increased (2.9 mm). On the basis of this measurement the estimated risk for fetal trisomy 21 was 1 : 317, rather than 1 : 934 on the basis of maternal age alone[1].

At the next ultrasound assessment at 22 weeks' gestation a large left sided pleural effusion was identified, with marked mediastinal shift resulting in the fetal heart lying entirely in the right half of the fetal chest. The remainder of the fetal anatomy appeared normal. The parents were informed of the possible risk of pulmonary hypoplasia and it was decided to insert a double pigtail pleuro-amniotic shunt. After the administration of local anesthesia a rigid trochar and cannula (De Ellis Medical Instruments, Croydon, UK) were introduced, under ultrasound guidance, through the maternal abdomen into the amniotic cavity and then through the fetal chest, in the left mid-axillary line, into the pleural cavity. The trochar was removed and the double pigtail catheter (KCH fetal bladder drainage catheter, Rocket Instruments, Watford, UK) was straightened and introduced into the cannula and, with a metal rod, one end of the catheter was deposited into the pleural cavity and the other end into the amniotic cavity[2,3].

At the time of shunt insertion, amniotic fluid was collected for fetal karyotyping and maternal blood was also taken to screen for infection; these examinations were subsequently reported as normal. Follow-up ultrasound examination at 24 weeks demonstrated resolution of the effusion, expansion of the lung and return of the heart to its normal position. Subsequent scans showed normal growth and activity of the fetus with no recurrence of the effusion.

In view of the past obstetric history an uneventful elective Cesarean section was carried out at 38 weeks' gestation and a boy weighing 3300 g was born in good condition. The shunt, which had been inserted in the mid-axillary line at the level of the fifth intercostal space, was clamped immediately at delivery to prevent development of a pneumo-thorax. The extra-fetal segment of the shunt lay looped around the fetal upper-arm, producing a circumferential constriction ring (Figures 1 and 2).

The baby did not require assisted ventilation and a plain chest radiograph demonstrated only a minimal pleural effusion. The shunt was removed but 24 h later he developed a pneumo-thorax that required the insertion of a chest drain. During the first 2 weeks of life there were recurrent pneumo-thoraces. Computerized tomography and magnetic resonance imaging showed no underlying lung pathology. Over the next 2 weeks the pneumo-thoraces resolved and the baby was discharged home in good condition.

Examination at delivery and subsequently over a period of several months demonstrated no impairment in the vascular supply to the limb or in its motor or sensory function and by 6 months of age there was only a faint ring around the upper limb.

DISCUSSION

This case illustrates a potentially dangerous complication

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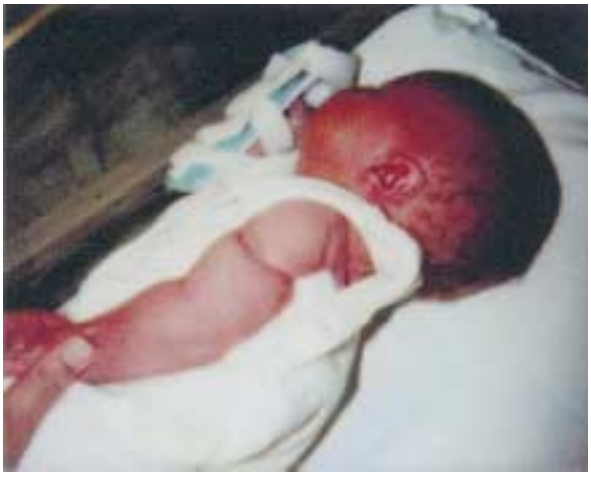

Figure 1 Constriction band around the upper arm at 48 h of age.

of the insertion of double pigtail fetal shunts with the entrapment of a limb.

There is a limited range of conditions for which insertion of an *in utero* shunt may be indicated including obstructive uropathy, pleural effusion and ventriculomegaly[4–6]. More usual complications of shunting include amniorrhexis, abortion, preterm labor or delivery and internalization or removal, presumably by the fetus, of the shunt[7,8]. Our policy in general with pleuro-amniotic shunts has therefore been to insert them in the dorsal region beneath the scapula, however, this is not always possible. Pigtail shunts are used as they reduce the likelihood for migration or internalization and as coiling of the extra-fetal portion of the shunt is thought to be less likely to result in entanglement. However in this case, the developing fetal limb was 'trapped' within this coiled segment with the development of a constriction band around the upper arm.

The amnion-disruption sequence or amniotic band syndrome although rare, is the most common cause of intrauterine constriction band formation. This sequence is associated with limb amputations, cranio-facial defects including clefting, visceral defects and fetal death[9–14]. The flexible nature of the shunt and its ability to unfold with growth of the enclosed fetal limb, would explain why entrapment of the limb was not associated with vascular impairment, and therefore either amputation or loss of function of the distal limb.

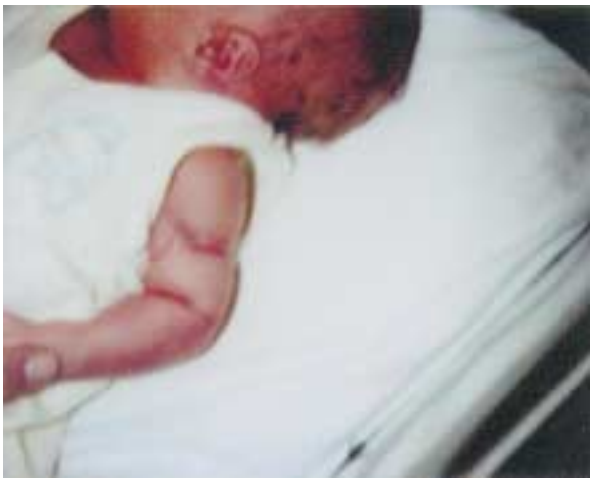

Figure 2 Constriction band around the infant's upper arm.

The lack of other similar cases in the literature would suggest either that this is a very rare complication of *in utero* shunting or that the complication has been unrecognized or unreported. The apparent rarity of this complication is reassuring, but it should nonetheless be borne in mind by those practitioners undertaking such procedures.

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