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Thoraco-Amniotic Shunting

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Abstract. Thoraco-amniotic shunting was performed in 51 singleton pregnancies for decompression and chronic drainage of fetal pleural effusions (n = 47), pericardial effusion (n = 1), or pulmonary cysts (n = 3). Five fetuses had chromosomal defects and in 4 the parents elected termination of pregnancy. All 18 non-hydropic fetuses and 14 of the 28 with hydrops survived. Thoraco-amniotic shunting is useful for diagnostic evaluation and treatment of fetuses with pathologic collection of intrathoracic fluid.

Introduction

Intra-uterine mediastinal compression by conditions such as cystic adenomatoid malformation and pleural effusions can lead to the development of hydrops and polyhydramnios which are associated with a high risk of premature delivery and intra-uterine or neonatal death. In previous studies we reported the outcome of 12 such fetuses treated by thoraco-amniotic shunting [1–3]. This paper reports data from our extended series of 51 cases and reviews the literature in this field.

Patients and Methods

During a 6-year period (1985–1990), thoraco-amniotic shunting was performed in 51 singleton pregnancies referred to our unit for investigation and management of fetal pleural effusions (n = 47), pericardial effusion (n = 1), or pulmonary cysts (n = 3). The diagnosis was made by ultrasonographic examination that was performed either because of polyhydramnios (n = 30) or as part of routine antenatal care (n = 21). Gestational age at referral was 19–35 (mean = 28) weeks. This was calculated from the maternal last menstrual period and confirmed by ultrasonographic measurement of fetal biparietal diameter.

In our centre an ultrasound scan was performed for the diagnosis of fetal malformations and assessment of the degree of associated skin oedema or ascites (fig. 1). Fetal karyotyping was performed by cordocentesis (n = 44) or amniocentesis (n = 7). After thoraco-amniotic shunting, serial ultrasound scans, mainly at the referring hospitals, were performed at weekly intervals to determine whether the effusions had reaccumulated. In these cases the patients returned to our unit for reassessment and insertion of new shunts when necessary. If the pregnancy continued labour and delivery proceeded without modification of normal clinical practice. After delivery the chest drains were immediately clamped and removed to avoid development of pneumothoraces.

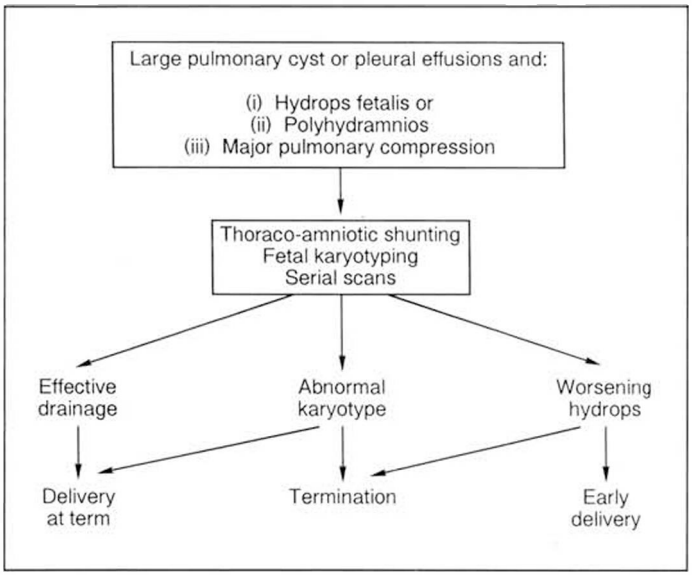


Fig. 1. Proposed management of abnormal accumulations of intrathoracic fluid. Thoraco-amniotic shunting should be consisdered if there is associated hydrops, polyhydramnios or major, progressive pulmonary compression and/or mediastinal shift. If after shunting the fetal karyotype is abnormal or there is worsening hydrops, the parents may choose to have termination of pregnancy. If there is reaccumulation of intrathoracic fluid further shunting may be undertaken.

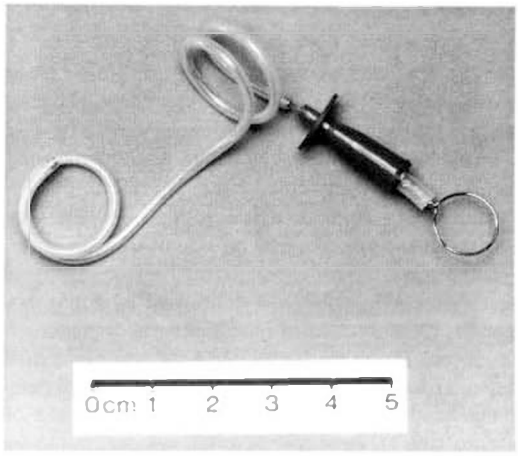


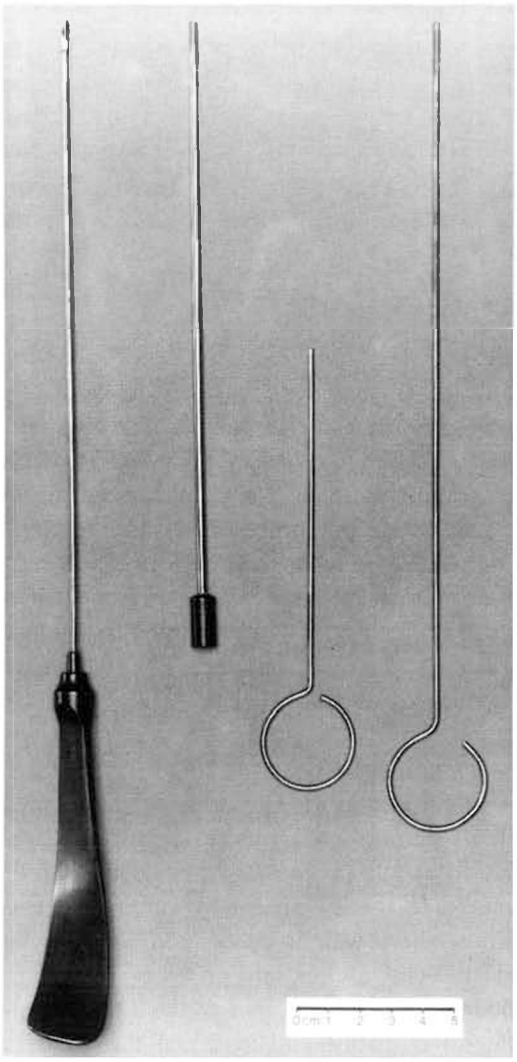
Fig. 2. Double pigtail catheter.

During the same 6-year period we examined a further 164 cases with hydrops and 39 with cystic adenomatoid malformation (microcystic = 20, macrocystic = 19). In these cases thoraco-amniotic shunting was not performed either because: (1) there were associated ultrasonically detectable lethal abnormalities; (2) the intrathoracic accumulation of fluid was not large enough to produce major pulmonary compression or mediastinal shift, or (3) the parents elected termination of pregnancy.

Technique of Thoraco-Amniotic Shunting

The effusions or cysts were drained into the amniotic cavity through a double pigtail silastic catheter (fig. 2; Rocket-Kings Catheters). The catheter, with external and internal diameters of 0.21 and 0.15 mm, respectively, has radiopaque stainless steel inserts at each end and lateral holes around the coil.

Shunting was performed as an out-patient procedure, without maternal sedation or fetal paralysis [1]. Ultrasound scanning, with a curvilinear transducer, was used to obtain a transverse section of the fetal thorax. With the transducer in one hand, held parallel to the intended course of the cannula, the chosen site of entry on the maternal abdomen was cleaned with antiseptic solution and local anaesthetic was injected down to the myometrium. Under ultrasound guidance, a metal cannula with a trochar (fig. 3; external diameter 3 mm, length 15 cm; RMS surgical developments) was introduced transabdominally into the amniotic cavity and inserted through the fetal chest wall, in the mid-thoracic region, into the effusion or cyst (fig. 4). The trochar was removed and in all cases



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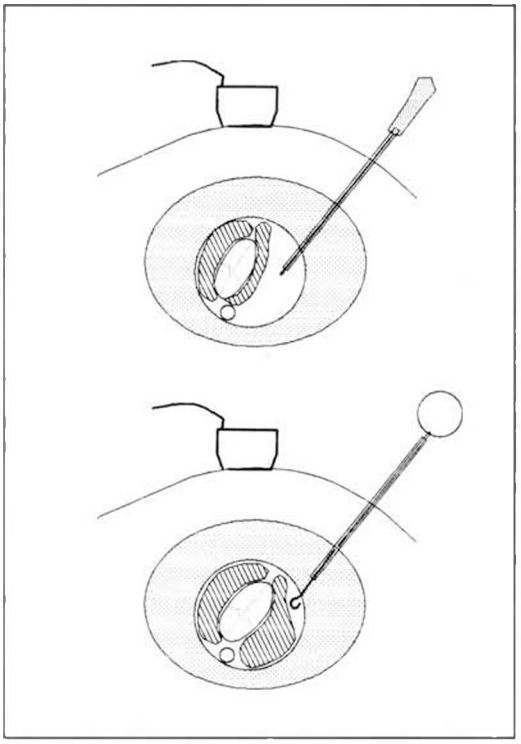

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Fig. 3. Trocar, cannula and rods used for the insertion of thoraco-amniotic shunts.

Fig. 4. For insertion of a thoraco-amniotic shunt a trochar and cannula are introduced through the mothers abdomen into the fetal thorax. The trochar is removed and a plastic catheter is threaded down the cannula. One end of the shunt is pushed into the fetal thorax. The cannula is withdrawn into the amniotic cavity where the outer end of the shunt is deposited.

straw-coloured fluid was drained. The catheter was then inserted into the cannula and a short introducer rod was used to deposit half of the catheter into the effusion or cyst. Subsequently, the cannula was gradually removed into the amniotic cavity where the other half of the catheter was pushed by a longer introducer. If drainage of the contralateral lung was also needed the appropriate fetal position was achieved by rotation of the fetal body using the tip of the cannula.

Results

Pericardial Effusion

A large fetal pericardial effusion with compression of both lungs was detected at routine ultrasound examination at 18 weeks of gestation (fig. 5). There were no cardiac or other malformations, the fetal karyotype was

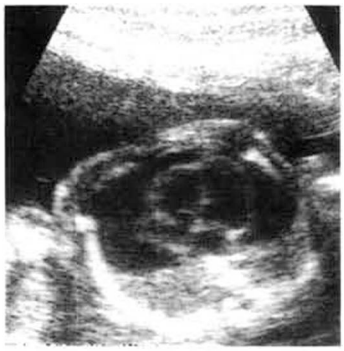

Fig. 5. Transverse section of the fetal thorax demonstrating lung compression due to a large pericardial effusion.

normal and an infection screen was negative. Ultrasound-guided needle aspiration of sterile, straw-coloured pericardial fluid was associated with rapid expansion of the lungs but the effusion reaccumulated within 48 h. At 21 weeks of gestation a catheter was introduced between the pericardial and amniotic cavities. Although at 25 weeks the catheter was found to be floating free in the amniotic cavity, there was no recurrence of the effusion. Subsequently, serial scans demonstrated normal fetal growth and a healthy infant was delivered vaginally after spontaneous onset of labour at term. Extensive investigations demonstrated normal cardiac anatomy and function and failed to identify a cause for the prenatal findings. Serial measurements of compliance of the respiratory system and functional residual capacity during the subsequent 12 months were normal.

Cystic Adenomatoid Malformation of the Lung

In three cases shunts were inserted at 24 [1], 25 and 32 weeks of gestation, respectively, for drainage of a large pulmonary cyst with hyperechogenic wall (fig. 6). The ultrasound appearances were compatible with cystic adenomatoid malformation of the lung (CAM type 1). Serial scans had previously demonstrated progressive enlargement of the cysts to occupy the whole hemithorax and causing mediastinal shift with compression of the contralateral lung.

Insertion of the shunts resulted in rapid expansion of the lungs. Follow-up scans showed no evidence of refilling of the cysts, and the pregnancies progressed uneventfully until spontaneous onset of labour and normal vaginal deliveries at 38, 39 and 33 weeks of gestation, respectively. During the subsequent 24–48 h thoracotomies and removal of the affected lobes were performed. In each case, histology revealed the presence of a large unilocular cyst, surrounded by smooth muscle and bars of cartilage. The infants are developing normally [4].

Pleural Effusions

Pleuro-amniotic shunting was performed in 47 fetuses with pleural effusions, diagnosed at 19–35 weeks of gestation (table 1; fig. 7). In the 22 cases with unilateral effusions, there was an associated mediastinal shift. Fetal ascites and/or generalised skin oedema was present in 8 of the 22 with unilateral effusions and in 22 of the 25 with bilateral effusions. Insertion of the shunts resulted in rapid expansion of the lungs in all but 1 case that was subsequently found to have arthrogryposis. In the cases with unilateral effusions, there was a simultaneous shift of the heart to its normal position in the thorax (fig. 8). In 4 cases the effusions reaccumulated 1–2 weeks after shunting, presumably because the fetus pulled out the shunt, and further shunts were inserted. Within 1–3 weeks after shunting, there was resolution of

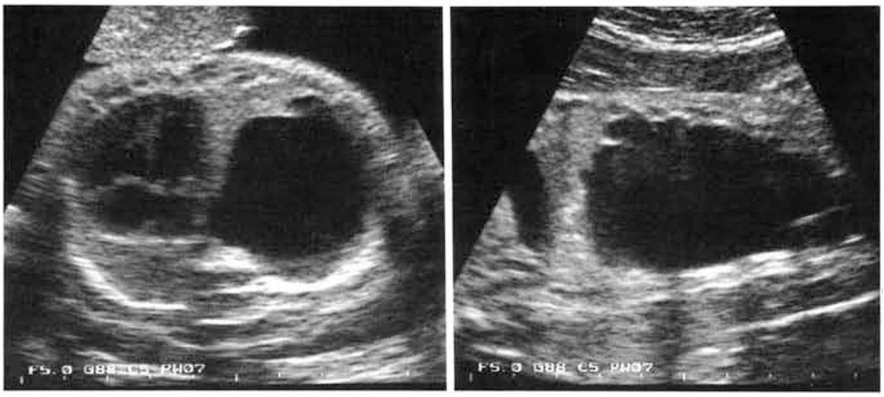

Fig. 6. Transverse (left) and longitudinal (right) section of the fetal thorax demonstrating lung compression and mediastinal shift due to a large cystic adenomatoid malformation (CAM type 1).

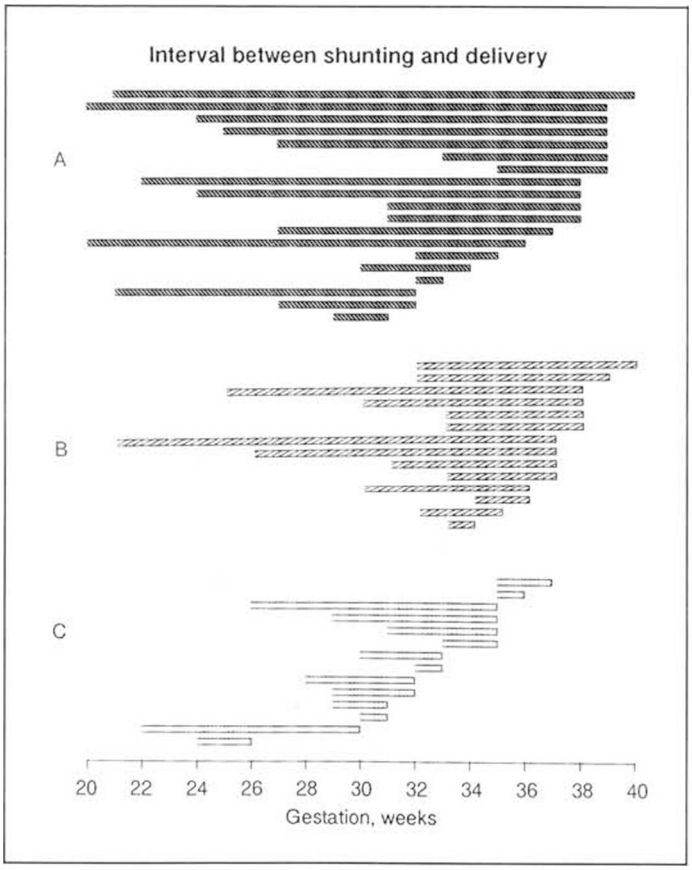

Fig. 7. Thoraco-amniotic shunting in 47 fetuses with pericardial effusion, cystic adenomatoid malformation or pleural effusions, including 30 with hydrops, treated at King's College Hospital 1985–1990. Four cases with fetal chromosomal defects where the pregnancies were terminated are not included. The horizontal lines connect the gestation at shunting with the gestation at delivery. In the non-hydropic group (**A**), all 19 infants survived. In the hydropic group 14 infants survived (**B**), and 14 died (**C**). Although the mean gestation at shunting was similar (A = 27, B = 30, C = 29 weeks), the time between shunting and delivery in group C was shorter (A = 10, B = 7, C = 3 weeks) presumably because in this group shunting did not prevent worsening of hydrops and/or development of polyhydramnios.

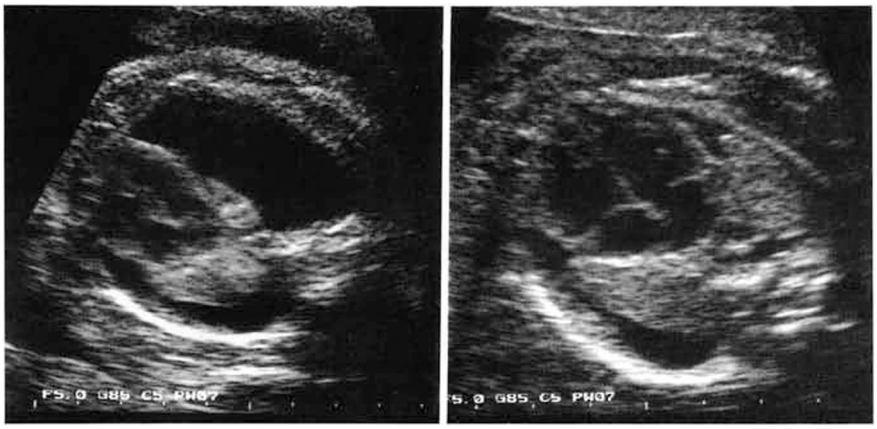

Fig. 8. Transverse section of the fetal thorax demonstrating large left pleural effusion and compression of both the heart and lungs (left). Immediately after thoraco-amniotic shunting (right) both lungs expanded and the heart returned to its normal position.

Table 1. Thoraco-amniotic shunting in 51 fetuses with pericardial effusion, cystic adenomatoid malformation or pleural effusions

Case No.	Pathology		Hydrops		AFV	Karyo-type	Gestation		Out-come	Comments
	site	type	A	E			TAS	delivery		
1	peric	E	–	–	N	46xx	21	40 V	alive	
2	L	CAM	–	–	N	46xx	24	38 V	alive	
3	R	CAM	–	–	N	46xy	25	39 El	alive	
4	L	CAM	–	–	P	46xy	32	33 V	alive	
5	L	PE	–	–	N	46xy	20	36 V	alive	
6	L	PE	–	–	N	46xy	20	39 V	alive	
7	R+L	PE	–	–	N	46xx	21	32 V	alive	
8	L	PE	–	–	N	46xy	22	38 El	alive	CDH
9	L	PE	–	–	P^R	46xy	24	39 V	alive	
10	L	PE	–	–	P	46xy	27	32 Em	alive	
11	R+L	PE	–	–	N	46xy	27	37 V	alive	
12	L	PE	–	–	N	46xx	27	39 V	alive	
13	R	PE	–	–	N	46xx	29	31 Em	alive	
14	L	PE	–	–	P^R	46xx	30	34 V	alive	
15	R	PE	–	–	P^R	46xx	31	38V	alive	
16	R	PE	–	–	N	46xy	31	38 V	alive	
17	R	PE	–	–	P^R	47xx+21	32	35 V	alive	AVSD
18	L	PE	–	–	P^R	46xy	33	39 V	alive	

Table 1 (continued)

Case No.	Pathology		Hydrops		AFV	Karyo-type	Gestation		Out-come	Comments
	site	type	A	E			TAS	delivery		
19	R+L	PE	–	–	P^R	46xx	35	39 V	alive	
20	R+L	PE	++	$++^R$	N	46xy	21	37 El	alive	
21	R+L	PE	++	$-^R$	N	46xy	25	38 V	alive	
22	R	PE	+	$+^R$	P^R	46xx	26	37 V	alive	
23	R+L	PE	+	$++^R$	P^R	46xx	30	36 V	alive	
24	R+L	PE	+	$+^R$	N	46xx	30+32	38 V	alive	
25	R+L	PE	++	$+^R$	P^R	46xy	31	37 V	alive	
26	R+L	PE	++	$+^R$	P^R	46xy	32	35 Em	alive	
27	L	PE	++	$-^R$	P^R	46xy	32	39 V	alive	
28	L	PE	+	$++^R$	P^R	46xx	32	40 V	alive	
29	R	PE	–	++	P	46xy	33	34 V	alive	thyroid teratoma
30	R+L	PE	++	$++^R$	P^R	46xy	33	37 Em	alive	
31	R	PE	++	$++^R$	P^R	46xy	33	38 V	alive	
32	R	PE	–	$++^R$	P^R	46xy	33	38 V	alive	
33	L	PE	++	++	P	46xy	34	36 V	alive	
34	R+L	PE	++	++	N	46xy	22	30 V	IUD	
35	R+L	PE	++	++	P^R	46xx	24	26 V	IUD	
36	R+L	PE	++	++	D	46xy	26	35 Em	NND	
37	R+L	PE	–	++	P^R	46xx	28	32 Em	NND	
38	R+L	PE	++	+	P^R	46xy	29+30	31 V	NND	
39	R+L	PE	++	++	P^R	46xx	29	32 Em	NND	
40	R+L	PE	++	++	P^R	46xx	29+32	35 V	NND	neonatal septicemia
41	R+L	PE	+	+	P	46xx	30	31 V	NND	
42	R+L	PE	++	++	P	46xy	30	33 V	NND	PH
43	R+L	PE	+	++	P	46xy	31+32	35 V	NND	arthrogryposis, PH
44	R+L	PE	–	++	P	46xy	32	33 V	NND	univentricular heart
45	R+L	PE	++	$+^R$	N	46xx	33	35 V	NND	CDH, PH
46	R	PE	+	++	P	46xy	35	37 V	NND	CDH, PH
47	R+L	PE	+	++	P	46xy	35	36 V	NND	
48	L	PE	–	–	N	45X/46XX	19	20 V	TOP	
49	L	PE	–	–	N	47xx+21	19	22 V	TOP	
50	R+L	PE	+	+	N	47xy+21	20	21 V	TOP	
51	R+L	PE	+	–	N	47xx+21	21	22 V	TOP	

Pathology site: L = left, R = right; Pathology type: PE = pleural effusion, CAM = cystic adenomatoid malformation; Hydrops: A = ascites, E = oedema, + = mild/moderate, ++ = severe, ^{R} = resolved; AFV = amniotic fluid volume, P = polyhydramnios, N = normal, D = decreased; TAS = thoraco-amniotic shunting; Delivery: V = vaginal, El = elective caesarean section, Em = emergency caesarean section; Outcome: TOP = termination of pregnancy, IUD = intra-uterine death, NND = neonatal death; Comments: AVSD = atrio-ventricular septal defect, CDH = congenital diaphragmatic hernia, PH = pulmonary hypoplasia.

the polyhydramnios in 20 of the 30 cases and the fetal hydrops resolved in 13 of the 28 affected pregnancies that continued.

Four pregnancies were terminated at the parents' request because the fetuses had trisomy 21 (n = 3) or Turner's mosaic (n = 1). Of the remaining 44 cases, all 15 non-hydropic fetuses survived. In the hydropic group (n = 28), 14 babies survived, 2 died in utero and 12 died in the neonatal period. The intra-uterine deaths, at 26 and 30 weeks of gestation, respectively, occurred 4–8 weeks after shunting which was effective in draining the pleural effusions but did not prevent progressive increase in skin oedema. The causes of neonatal death were: major cardiac defect (n = 1); arthrogryposis (n = 1); neonatal pseudomonas septicemia (n = 1); pulmonary hypoplasia secondary to an associated diaphragmatic hernia (n = 2), and in 7 cases a combination of respiratory, cardiovascular and renal failure of unidentified aetiology. In the 29 surviving infants (3 months to 6 years of age), there is normal growth and development with no respiratory morbidity.

Prognostic signs for poor outcome were the presence of associated malformations, bilateral rather than unilateral pleural effusions and the presence of hydrops fetalis and/or polyhydramnios that did not resolve within 1–3 weeks after shunting (table 1).

Literature Review

Cystic Adenomatoid Malformation of the Lung

Cystic adenomatoid malformation of the lung is a rare congenital abnormality with less than 200 cases reported in the literature. The aetiology is unknown, but the lack of differentiation of the respiratory structures involved suggests that it originates before the 5th week of embryological life [5]. There is a broad spectrum of clinical presentations. Some infants present in the first week of life with severe, and often fatal, respiratory insufficiency. Smaller lesions are often asymptomatic until late childhood and they are easily corrected by excision of the affected pulmonary segment or lobe.

Prenatal diagnosis is based on the ultrasonographic demonstration of a hyperechogenic pulmonary tumour which is either solid (microcystic) or cystic (type 1). Polyhydramnios is a common feature and this may be a consequence of decreased fetal swallowing of amniotic fluid due to oesophageal compression, or increased fluid production by the abnormal lung tissue. When there is compression of the heart and major blood vessels in the thorax fetal hydrops develops.

Prognostic features for poor outcome include microcystic disease, major lung compression causing pulmonary hypoplasia, and development of hydrops fetalis irrespective of the type of lesion. Azdick [6] has recently examined 18 cases and reviewed another 17 from the literature. In 14 of the 15 cases with microcystic disease the fetuses were hydropic and they all died either before or after birth; the 1 non-hydropic fetus survived. In contrast, 16 of the 22 cases with macrocystic disease were non-hydropic and all but 1 survived; none of the 6 cases with hydrops survived.

There are 2 cases reported where thoracoamniotic shunting was performed at 20 and 24 weeks of gestation, respectively, for decompression of an intrathoracic cyst either because of associated hydrops or progressive enlargement of the cyst and major mediasti-

nal shift [1, 7]. Insertion of the shunts resulted in rapid expansion of the lungs and gradual resolution of the hydrops. Serial scans showed no evidence of refilling of the cysts and the pregnancies progressed uneventfully. The infants were delivered in good condition at term and had successful postnatal surgery. Subsequent growth and development were normal.

Pleural Effusions

Hydrops fetalis, with an incidence of 3–10 per 10,000 births, is characterised by generalised skin oedema and pericardial, pleural, or ascitic effusions. This is a non-specific finding in a wide variety of fetal and maternal disorders, including haematological, chromosomal, cardiovascular, pulmonary, gastrointestinal, hepatic and metabolic abnormalities, congenital infection, neoplasms and malformations of the placenta or umbilical cord. While in many instances the underlying cause may be determined by detailed ultrasound scanning and fetal blood sampling, frequently the abnormality remains unexplained even after expert postmortem examination [8–10].

Fetal pleural effusions may be an isolated finding or they occur in association with generalised oedema and ascites. Irrespective of the underlying cause, infants affected by pleural effusions usually present in the neonatal period with severe, and often fatal, respiratory insufficiency. This is either a direct result of pulmonary compression caused by the effusions, or due to pulmonary hypoplasia secondary to chronic intrathoracic compression. The overall mortality of neonates with pleural effusions is 25%, with a range from 15% in infants with isolated pleural effusions to 95% in those with gross hydrops [11]. More recently, Longaker et al. [12] reported that the mortality rate in cases of antenatally diagnosed chylothorax was 53%.

Spontaneous Resolution

There are at least 3 case reports of fetuses with isolated pleural effusions, diagnosed at 16, 25 and 32 weeks of gestation, where spontaneous resolution was documented within 2–7 weeks [13–15]. Furthermore, Longaker et al. [12] and Pijpers et al. [16] examined the natural history of antenatally diagnosed pleural effusions in 24 and 8 cases, respectively, and reported spontaneous resolution in a total of 5 fetuses.

In contrast, there are at least 5 reported cases with isolated pleural effusions, diagnosed at 30–36 weeks of gestation, where no antenatal drainage was performed and the outcome was either intra-uterine or early neonatal death due to pulmonary hypoplasia [17–21]. Furthermore, Benacerraf et al. [22] reported a case with isolated pleural effusions at 20 weeks where the fetus subsequently became hydropic and died.

Neonatal Thoracocentesis

Lange and Manning [23], Bruno et al. [24] and Pijpers et al. [16] reported the diagnosis of isolated pleural effusions in a total of 9 fetuses at 25–39 weeks of gestation that were successfully drained immediately after delivery and all babies survived. In contrast, in the series of Longaker et al. [12] only 10 of 20 infants requiring postnatal drainage of the effusions survived.

Intrapartum Thoracocentesis

Petres et al. [25] drained the right pleural effusion of a fetus at 36 weeks of gestation, but this reaccumulated within 24 h. At 37 weeks, when the mother was in labour, the

effusion was drained and the infant was born in good condition. Similarly, Schmidt et al. [26] reported the successful intrapartum thoracocentesis and paracentesis in a hydropic fetus at 35 weeks of gestation; the infant required assisted ventilation until the 20th day after birth and survived.

Intra-Uterine Thoracocentesis or Short-Term Drainage

Kurjak et al. [27] and Benacerraf et al. [22] reported 3 cases with isolated pleural effusions at 19–22 weeks of gestation. Repeated thoracocenteses were performed every few days until 24 weeks when the effusions resolved spontaneously. The healthy infants were delivered at term and did not require any postnatal therapy.

Similarly good results were reported by Roberts et al. [28] who inserted one end of an epidural catheter into the unilateral pleural effusion of a fetus at 25 weeks of gestation; the other end of the catheter was placed in a bag on the mother's abdomen. During the first 24 h, 70 ml of bright yellow fluid was drained, but only 30 ml drained over the next 5 days and subsequently the catheter was removed. The effusion did not reaccumulate and a healthy infant was delivered at term.

In contrast, Meizner [29], Longaker et al. [12] and Landy et al. [30] performed thoracocenteses at 28–34 weeks of gestation in 6 fetuses with pleural effusions, including 2 with hydrops. In all cases there was rapid reaccumulation of the effusions and the infants, delivered within 4 weeks of diagnosis, died soon after birth due to pulmonary insufficiency.

Thoraco-Amniotic Shunting

Seeds and Bowes [31] inserted thoraco-amniotic catheters in a fetus with large pleural effusions at 30 weeks of gestation. Postoperatively, fetal activity increased, polyhydramnios decreased and there was cessation of the uterine contractions that were present before shunting. However, after 3 days the effusions reaccumulated because one of the catheters had come out of the fetal chest, and the other catheter had been drawn under the fetal skin surface. Polyhydramnios increased and the infant, delivered 48 h later, made a good recovery after drainage and ventilatory support.

Weiner et al. [32] performed thoraco-amniotic shunting at 24 weeks in a hydropic fetus with extralobar sequestration. Although there was resolution of the oedema and ascites, the infant born at 29 weeks died in the neonatal period.

Rodeck et al. [33] reported 8 cases with pleural effusions, including 5 with hydrops, that were shunted 25–35 weeks of gestation. The infants were delivered at 32–39 weeks of gestation; 6 infants survived with good respiratory function and 2 died in the neonatal period due to pulmonary hypoplasia. In the latter 2 cases the lungs did not expand after thoraco-amniotic shunting.

Conclusions

Isolated pleural and pericardial effusions or pulmonary cysts in the fetus may either resolve spontaneously or they can be treated effectively after birth. Nevertheless, in some cases severe and chronic compression of the fetal lungs can result in pulmonary hypoplasia and neonatal death. In others, mediastinal compression leads to the development of hydrops and polyhydramnios which are associated with a high risk of premature delivery and perinatal death.

The data from fetuses with isolated pleural or pericardial effusions suggest that certainly in some cases short-term decompression by thoracocenteses or temporary drainage may disrupt the underlying pathology. However, in the majority of cases the fluid reaccumulates within 24 h requiring repeated procedures which are likely to be more traumatic than thoraco-amniotic shunting.

The data from the present study indicate that thoraco-amniotic shunting is useful for the diagnostic evaluation of fetuses with pathologic collection of intrathoracic fluid. Firstly, the diagnosis of an underlying cardiac abnormality or other intrathoracic lesion may become apparent only after effective decompression and return of the mediastinum to its normal position. Secondly, it may be useful in the prenatal diagnosis of pulmonary hypoplasia because in such cases the lungs often fail to expand after shunting. Thirdly, it may help distinguish between hydrops due to primary accumulation of pleural effusions, in which case the ascites and skin oedema may resolve after shunting, and other causes of hydrops such as infection, in which drainage of the effusions does not prevent worsening of the hydrops.

Thoraco-amniotic shunting is also an effective and apparently safe method of chronic drainage of fetal pleural effusions or pulmonary cysts. It can reverse fetal hydrops, resolve polyhydramnios and thereby reduce the risk of preterm delivery, and may prevent pulmonary hypoplasia. The alternative management of pleural effusions by thoracocentesis, immediately before or after delivery, could prevent respiratory distress in the neonate if this is the result of simple mechanical compression of the otherwise normally developed lung. However, such treatment would not prevent pulmonary hypoplasia, due to prolonged intrathoracic compression, or indeed progressive disease from pleural effusions to hydrops fetalis and intra-uterine death. Nevertheless, in a high proportion of hydropic fetuses (50% in our series), thoraco-amniotic shunting does not prevent their ultimate death due to the underlying disease causing the hydrops.

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