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Respiratory Function in Infancy following Pleuro-Amniotic Shunting

Key Words
Antenatal diagnosis
Lung function
Pleural effusion

Abstract
Respiratory status was assessed at a median of 12 (range 3–60) months in 17 infants who had undergone pleuro-amniotic shunting to drain fetal pleural effusions. The shunts had been inserted at a median of 29 weeks (range 21–35) gestation; 12 fetuses were hydropic at the time of shunting. In all 17 cases effective chronic drainage of the pleural effusions was achieved. Respiratory status at follow-up was assessed by documentation of respiratory symptoms and measurement of functional residual capacity (FRC) by a helium gas dilution technique. Six infants suffered from recurrent respiratory symptoms, this incidence (37%) did not differ significantly from that found in a control group. The mean FRC of the study population was 28 ml/kg (range 19–34 ml/kg), only 2 infants' FRCs were below the reference range. These results suggest that pleuro-amniotic shunting may, by effective drainage of pleural effusion and hence prevention of chronic antenatal intrathoracic compression, avoid impairment of antenatal lung growth.

Introduction

Prolonged intrathoracic compression of the fetal lung may result in pulmonary hypoplasia and neonatal death [1–3]. Certain conditions which result in intrathoracic compression are amenable to antenatal therapy [4–6]. Pleuro-amniotic shunting [5, 6] is an effective and apparently safe method of achieving chronic drainage of fetal pleural effusions [5]. In one series [6] none of 11 infants so treated had fatal respiratory problems in the neonatal

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period, suggesting that pleuro-amniotic shunting had prevented serious impairment of antenatal lung growth. Pulmonary hypoplasia, however, does not always result in neonatal death [7]. Thus assessment of the respiratory status of survivors is essential if the effectiveness of antenatal interventions aimed at relieving pulmonary compression is to be accurately determined. This study reports the respiratory follow-up of infants who had undergone antenatal pleuro-amniotic shunting to achieve drainage of fetal pleural effusions.

Patients and Methods

During a 6-year period (1985–1990) 48 fetuses with pleural effusions had pleuro-amniotic shunts inserted in our unit [8]. Twenty-eight infants survived the neonatal period, but 1 infant subsequently died at 6 months of age of septicaemia, she had severe necrotising enterocolitis in the neonatal period and required prolonged intravenous nutrition. Seventeen of the 27 survivors lived close enough to King's College Hospital to attend for respiratory follow-up and formed the study group. There was no significant difference in the gestational age at insertion of the pleuro-amniotic shunts between this group (mean 29 weeks, range 21–35) and the 10 survivors not followed-up (mean 29 weeks, range 20–34), or in their gestational age at delivery (mean 37 weeks, range 31–40, and mean 36 weeks, range 31–39, respectively).

Eight of the 17 patients of the study group had had bilateral pleuro-amniotic shunts inserted and 12 fetuses were hydropic at the time of shunting (cases 1–12, table 1). In all cases insertion of the pleuro-amniotic shunt resulted in immediate drainage of the effusion with associated lung expansion and subsequent resolution of the hydrops in the 12 affected cases. In the 9 fetuses with unilateral effusions, drainage of the effusion resulted in return of the mediastinum to the midline. In only 1 infant (case 1, table 1) did the pleural effusion subsequently reaccumulate and this was due to presumed removal of the shunt by the fetus. A further drain was inserted which achieved chronic drainage for the 6 weeks until delivery.

This study was approved by the King's College Hospital Ethics Committee.

The infants were seen in the Paediatric Respiratory Laboratory at a median of 12 months of age (range 3–60 months). A clinical history was taken and the infants were weighed and examined. Parents were asked about the occurrence of their infant's respiratory symptoms. The infants were diagnosed to be suffering from recurrent respiratory symptoms if they wheezed or coughed on at least 3 days/week for a 4-week period or if these symptoms were present for at least 2 days following all upper respiratory tract infections [9]. Parents were also asked to give details of any medication prescribed for their child, in particular if this included bronchodilator therapy or antibiotics for chest infections. Data obtained from parents were then subsequently confirmed by examination of the hospital records.

Lung function was assessed by measurement of function residual capacity (FRC) using a helium gas dilution technique. All measurements were made with the infant in the semi-prone position. The infant breathed through a face mask, held firmly in place to prevent leaks, into a water-sealed spirometer (Gould Pulmonet III). The accuracy of the spirometer was checked daily with calibrated syringes. The spirometer incorporates a digital display of FRC which was recorded at 15-second intervals. When the display remained unchanged for 30 s, equilibration was assumed to have occurred and the measurement discontinued. The traces were coded and analysed blind of clinical details by A.G. From the trace the end expiratory level was determined and FRC calculated. The results were converted to body temperature, pressure saturated conditions and related to the infant's body weight.

To assess the reproducibility of the measurement of FRC in young infants, two separate measurements were made in 30 children with a similar postnatal age to the study population. The mean of the differences between the paired measurements was 1.8 ml/kg. The intra-subject reproducibility of the measurement in infants and young children had been previously calculated to be 7.3%. The mean FRC of 50 healthy infants (controls) measured in our own laboratory of postnatal age range 0–2 years was 30 ml/kg (95% confidence limits ± 6 ml/kg). The FRC of children older than 2 years of age was compared with published normal data [10], their results were considered abnormal if they were above 120% or below 80% of that predicted for age [11].

Statistical Analysis

Differences between the groups were assessed for statistical significance using either the Wilcoxon rank sum test or Fisher's exact test. To assess whether the

Table 1. Individual infants' characteristics

Case No.	Antenatal					Postnatal		
	GA	GE	pleural effusion	ascites	oedema	age	FRC ml/kg	resp. symptoms
1	30	38	LR	+	+	12	27	No
2	33	37	LR	+	+	6	28	No
3	31	37	LR	+	+	24	28	No
4	30	36	LR	+	+	18	23	No
5	21	37	LR	+	+	10	32	Yes
6	25	38	LR	+	+	4	29	Yes
7	32	40	L	+		5	31	Yes
8	33	38	R	+	+	12	25	No
9	26	37	R	+	+	15	28	Yes
10	33	34	R		+	33	32[a]	Yes
11	33	38	R		+	18	27	Yes
12	32	39	L	+		3	27	No
13	21	32	LR			10	19	No
14	35	40	LR			4	34	No
15	22	38	L			6	32	No
16	28	31	L			60	34[b]	No
17	31	38	R			14	25	No

GA = Gestational age (weeks) at pleuro-amniotic shunting; GE = gestational age (weeks) at delivery; L = left; R = right; + = present; Age = postnatal age (months) at examination; FRC = functional residual capacity.

[a] 84% of that predicted for age.

[b] 92% of that predicted for age.

relationship of FRC to gestational age at insertion of shunt was significant, a Spearman's correlation coefficient was calculated.

Results

No infant was symptomatic at the time of examination, although 6 infants were reported by their parents to have previously had recurrent respiratory symptoms. Four of the 6 infants were receiving regular bronchodilator medication and the other 2 had received frequent courses of antibiotics. Two of the symptomatic infants had had bilateral pleuro-amniotic shunts inserted and 4 had had unilateral shunts (non-significant). All of the symptomatic infants had been hydropic prior to insertion of the pleuro-amniotic shunt ($p = 0.145$; table 1).

The mean FRC of the study group was 28 ml/kg (range 19–34 ml/kg). Fifteen of the patients had FRCs within the normal ranges. The remaining 2 infants had low FRCs (cases 4 and 13, table 1), neither infant had recurrent symptoms. These two infants did not differ significantly from the rest of the study group in the gestational age at insertion of the pleuro-amniotic shunt, only 1 of the infants had been hydropic at the time of shunting.

There was no statistically significant difference between the FRC of the infants who

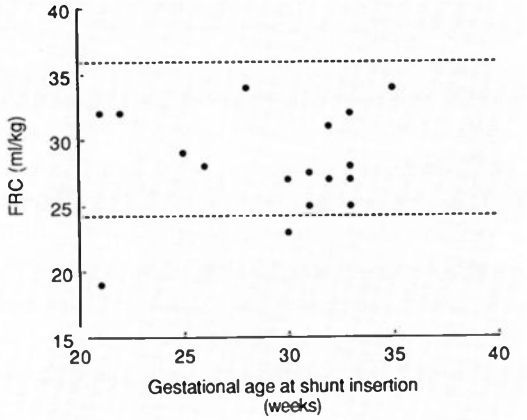

Fig. 1. Relationship of FRC to gestational age at insertion of the pleuro-amniotic shunts.

Table 2. Relationship of FRC to antenatal and postnatal problems

	FRC, ml/kg	
	median	range
Antenatal problems		
Bilateral shunt	28	19–34
Unilateral shunt	28	25–34
Hydropic	28	23–32
Non-hydropic	32	19–34
Postnatal problems		
Symptomatic	30	27–32
Non-symptomatic	27	19–34

required unilateral or bilateral drains, had or had not been hydropic at the time of insertion of the shunt or who were symptomatic or asymptomatic at follow-up (table 2). No significant correlation was documented between FRC and the gestational age at insertion of the pleuro-amniotic shunt ($r^2 = 0.011$; fig. 1).

Discussion

The present data suggest that pleuro-amniotic shunting may prevent the serious respiratory morbidity that has previously been documented following chronic antenatal intrathoracic compression [12]. Only 6 of the 17 infants (36%) we followed were symptomatic during the follow-up period. This incidence is similar to that found in an unselected group of infants born at term [13], yet some of our study population were delivered prematurely and the incidence of recurrent respiratory symptoms in preterm infants can be as high as 60% [13].

The mean FRC of the study population was within the reference range, suggesting that pleuro-amniotic shunting may have permitted normal antenatal lung growth in the majority of infants. Infants with impaired lung growth, pulmonary hypoplasia, have previously been documented to have small-volume lungs [14]. Lung volume may be assessed by measurement of thoracic gas volume (TGV) using a plethysmographic technique or by measurement of functional residual capacity. Unfortunately measurements of TGV could not be made on all our study population, some of whom were too old to be sedated and yet too young to co-operate with the manoeuvres necessary for TGV estimation. In contrast, FRC may be measured without sedation and requires no co-operation. Thus measurement of FRC could be applied to all the study population, facilitating comparison of patients.

Two infants had low FRCs, but one result was just below the 95% confidence limit of the reference range. It is difficult to explain why 1 infant had a very low FRC. The infant did not differ from the rest of the cohort regarding gestational age at insertion of the pleuro-amniotic shunts and other infants had suffered from bilateral effusions but had nor-

mal FRCs at follow-up. This infant will be remeasured at a later date. Hopefully, the infant's lung volume will then have increased to be with in the normal range, this has been seen in some infants who initially had low lung volumes following premature rupture of the membranes [15] and perhaps indicates catch-up lung growth postnatally.

The infants with recurrent symptoms did not have significantly different FRCs from infants without chronic symtoms. This is, however, not surprising, as none of the infants were symptomatic at the time of study [16] and all 6 had previously received or were receiving appropriate therapy [9].

Twelve of the 17 fetuses were hydropic prior to insertion of the pleuro-amniotic shunts. Untreated, this group would be expected to have a poor outcome [17]. In one series [18] no hydropic fetus with chronic pleural effusions survived. In the present series, although 6 of the infants previously hydropic had had recurrent symptoms, none had an abnormal FRC. These data suggest that pleuro-amniotic shunting may have been responsible for this improved outcome.

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