Impaired Respiratory Function in Infants With Anterior Abdominal Wall Defects

By P.J. Thompson, A. Greenough, E. Dykes, and K.H. Nicolaides
London, England

● **Respiratory function at follow-up was assessed in 13 infants who had undergone surgical repair of an anterior abdominal wall defect. Six infants had exomphalos and seven had gastroschisis. The infants were delivered at a median gestational age of 36 weeks (range, 32 to 38 weeks). Respiratory function was assessed by measurement of functional residual capacity (FRC) at a median postnatal age of 5 months (range, 1 to 10 months). Although there was no significant difference in the FRC of the infants with gastroschisis compared with those with exomphalos, the study group's mean FRC (25 mL/kg) was significantly lower than that of 50 healthy control infants (mean, 30 mL/kg; 95% CI, ±6 mL/kg). Five infants had an FRC below the 95% confidence limit of the normal range. These data suggest that infants with anterior abdominal wall defects may have impaired antenatal lung growth.**

INDEX WORDS: Exomphalos, gastroschisis, lung function.

ANTERIOR ABDOMINAL wall defects are relatively common with an incidence of approximately 1 in 3,500 live births.[1] One cause of neonatal mortality among these infants is respiratory insufficiency, which is usually ascribed to increased intraabdominal pressure following surgical repair.[2-5] This increase in intraabdominal pressure is limited to the immediate postoperative period and, thus, has only a temporary effect on lung function. There is, however, some evidence to suggest that respiratory insufficiency in infants with abdominal wall defects could also be due to impaired antenatal lung growth. Griscom and Driscoll reported stillbirths with exomphalos have small chests.[6] Furthermore, Hershenson et al[7] examined the chest radiographs of infants with giant exomphalos and found them to have reduced chest widths and lung areas. Impaired antenatal lung growth results in chronic abnormalities of respiratory function.[8] Therefore, we examined lung function of infants with anterior abdominal wall defects in the postneonatal period (this timing was chosen to avoid any acute effects on lung function resulting from the surgical intervention) to determine if such infants do indeed suffer from impaired antenatal lung growth.

MATERIALS AND METHODS

During an 18-month period (March 1990 to August 1991), 22 infants with anterior abdominal wall defects were delivered at King's College Hospital (KCH). All patients were referred to our unit antenatally for confirmation of the diagnosis of exomphalos (n = 10) or gastroschisis (n = 12), or exclusion of associated anomalies. Fifteen infants were delivered vaginally, after spontaneous (n = 7) or induced (n = 8) labor and seven were delivered by elective (n = 2) or emergency (n = 5) cesarean section for fetal distress in labor. Four infants, all with exomphalos, died before leaving the hospital. One infant died of septicemia related to a central feeding line; another developed a volvulus postoperatively with necrosis of the small bowel. The remaining two infants had multiple congenital anomalies including cardiac defects; both died of respiratory failure.

Thirteen of the 18 survivors attended for postnatal respiratory follow-up and formed the study group. There was no significant difference in these 13 infants from the rest of the cohort with respect to gestational age at delivery or requirement for respiratory support, or between the infants with gastroschisis or exomphalos who were followed-up (Table 1).

The infants were seen in the pediatric respiratory laboratory at a median of 5 months (range, 1 to 10). The infants were weighed and examined; their parents were asked about the nature of any current medication and if their child suffered from respiratory symptoms. Infants were described as being symptomatic if they had wheezed and/or coughed on at least 2 days per week over the previous month.[9]

Lung function was then assessed by measurement of functional residual capacity (FRC) using a helium gas dilution technique. All measurements were made with the infant in the semiprone position. The infants breathed through a face mask, held firmly in place to prevent air leaks, into a water-sealed spirometer (Gould Pulmonet III; Ilford, England). The accuracy of the spirometer was checked daily with calibrated syringes. The spirometer incorporates a digital display of FRC, which was recorded above the respiratory trace at 15-second intervals. When the display remained unchanged for 30 seconds equilibration was assumed to have occurred and the measurement was discontinued. The traces were coded and analyzed blind of clinical details by one of the authors (A.G.). From the trace the end expiratory level was determined and from this the FRC calculated. The results were converted to body temperature and pressure-saturated conditions and then related to the infant's 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 intrasubject reproducibility of the measurement in infants and young children had been previously calculated to be 7.3%.

FRC was also measured in 50 healthy infants (controls) all born between 37 and 41 weeks gestational age. The infants were of a

From the Department of Child Health and the Harris Birthright Research Centre for Fetal Medicine, King's College Hospital, London, England.
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Address reprint requests to A. Greenough, MD, Department of Child Health, King's College Hospital, London SE5 9RX, England.

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Table 1. Median and Range of Birth Weight, Gestation at Delivery, and Neonatal Respiratory Support in the Infants With Anterior Abdominal Wall Defects

	All Patients (n = 22)	Study Group	
		Gastroschisis (n = 7)	Exomphalos (n = 6)
Birthweight (g)	2,439 (1,262-3,278)	2,268 (1,732-3,182)	2,863 (2,010-3,278)
Gestational age (wk)	36 (29-38)	36 (34-37)	38 (32-38)
Duration of ventilation (d)	6 (1-64)	5 (1-17)	8 (1-21)
Maximum peak pressure (cm H_2O)	22 (16-42)	22 (17-30)	23 (18-26)
Maximum inspired oxygen (%)	54 (21-100)	70 (21-100)	48 (21-70)

similar postnatal age range (1 to 10 months) to the study population and their mean FRC was 30 mL/kg (95% CI ± 6 mL/kg).

Statistical Analysis

Differences between the study and the control group were assessed for statistical significance using the Student's *t* test. The confidence intervals were calculated with the appropriate *P* value from the standard error of the difference between the means of the two groups. Differences between the infants with gastroschisis and exomphalos were assessed for statistical significance using the Wilcoxon rank-sum test.

RESULTS

None of the infants was symptomatic or receiving regular medication at the time of examination. The mean FRC of the study group was 25 mL/kg (range, 18 to 31 mL/kg), which was significantly lower than that of the control group ($P < .01$) (Fig 1). The 95% confidence intervals of the differences of the means of the study group and the controls were 2.7 to 6.9 mL/kg. There was no significant difference in the FRCs of those infants with exomphalos (median FRC, 26; range, 21 to 30 mL/kg) from those with gastroschisis (median FRC, 26; range, 18 to 32 mL/kg).

No infant had an FRC above the 95% confidence limit of the control data, but five had an FRC below this, three with gastroschisis and two with exomphalos. These five infants did not differ significantly from the rest of the group with respect to gestational age at delivery, length of respiratory support, or postnatal age at follow-up (Table 2).

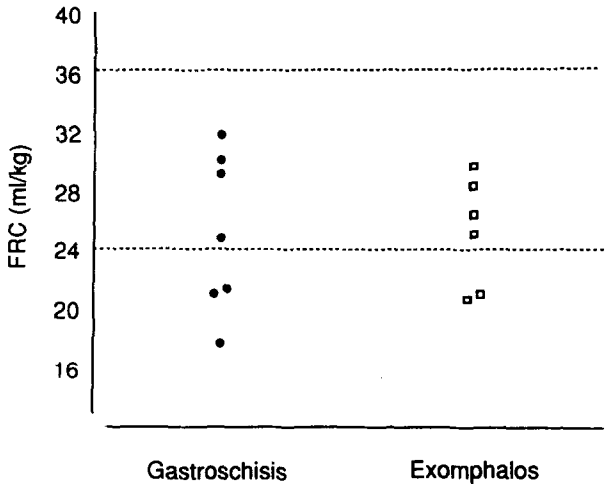

Fig 1. FRC results of the infants with gastroschisis and exomphalos (individual data demonstrated). The broken lines represent the 95% confidence limits of the control data.

DISCUSSION

This study demonstrates that postneonatal FRC is reduced in some infants with anterior abdominal wall defects. FRC is an assessment of lung volume. Infants with impaired antenatal lung growth have small volume lungs.[10] Thus, these results suggest that certain infants with anterior abdominal wall defects have impairment of antenatal lung growth and development.

FRC was assessed by a helium gas dilution technique. This method of assessment of lung volume may be inaccurate in infants with high airways resistance. It is unlikely, however, that any of the infants suffered from that abnormality of lung function as none were symptomatic[9] and infants with impaired antenatal lung growth have low rather than high airways resistance.[8]

Table 2. Gestational Age at Delivery, Neonatal Respiratory Support, and FRC at Follow-Up in Infants With Gastroschisis or Exomphalos

		Neonatal Respiratory Support				
Gestational Age (wk)	Diagnosis	Max PIP (cm H_2O)	Max FiO_2	Duration IPPV (d)	Age at Follow-Up (mos)	FRC (mL/kg)
35	G	21	70	6	2	18
36	G	22	70	7	2	21
36	G	24	40	1	5	21
37	G	22	80	5	7	25
36	G	17	21	1	5	29
35	G	30	100	17	7	30
34	G	18	21	2	1	31
38	E	25	40	5	10	21
38	E	18	21	1	2	21
38	E	20	45	5	8	25
32	E	26	50	21	1	26
37	E	22	70	13	10	28
38	E	26	54	11	9	30

Abbreviations: PIP, peak inspiratory pressure; IPPV, intermittent positive-pressure ventilation; G, gastroschisis; E, exomphalos.

One possible explanation for the observed reduction in lung volume is external compression of the lungs. This is an invariable finding in the immediate postoperative period, as restoration of the bowel into the abdominal cavity and closure of the anterior abdominal wall results in increased intraabdominal pressure with upward displacement of the diaphragm. However, this causes acute rather than chronic respiratory insufficiency because, as the infant grows, the intraabdominal pressure decreases. Therefore, the low postnatal FRCs of the study group are likely to be a consequence of impaired antenatal lung growth rather than an effect of the surgical intervention.

There are a number of causes of impaired antenatal lung growth including chronic pulmonary compression and cessation of fetal breathing activity. In utero, the majority of the abdominal contents of infants with gastroschisis or exomphalos are outside the abdominal cavity. The reduction in abdominal viscera in the upper part of the abdominal cavity may provide an inadequate framework for chest wall development which would restrict lung growth by external compression. This hypothesis is supported by the finding of reduced chest widths and lung areas on the chest radiographs of infants with giant exomphalos.[7] In addition, fetal breathing may be poor in these infants, the low intraabdominal pressure impairing diaphragmatic function. Diaphragmatic defects, both hernia and eventration, are common in association with anterior abdominal wall defects[7,11]; these conditions may represent the severe end of a spectrum of diaphragmatic abnormalities. Normal diaphragmatic function in utero is essential for normal lung development. Damage to the phrenic nerve[12] and abnormal diaphragmatic development, such as accessory hemidiaphragm,[13,14] have both been associated with impaired lung growth.

Previous studies have suggested that exomphalos compared to gastroschisis is associated with greater respiratory impairment, but this has only been documented in the neonatal period. Infants with exomphalos require longer ventilator support and have smaller chests documented by their chest radiographs.[7,15,16] We did not find any difference in the FRC results of the two groups at follow-up, but four infants (all with exomphalos) had died in the neonatal period and this may have influenced our results. If our proposed mechanisms of impaired antenatal lung growth in infants with anterior abdominal wall defects are correct, the reduction in FRC would correlate with the size of extraabdominal organs in utero and not the type of defect. We hope to test this hypothesis as greater numbers of infants are recruited into our prospective follow-up study.

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