Respiratory function in childhood following repair of oesophageal atresia and tracheoesophageal fistula

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Abstract

Aim—To determine the relation between respiratory function in infancy and at school age in children who have undergone oesophageal atresia and tracheoesophageal fistula repair, and assess the value of infant respiratory function testing; and to examine the effect of bronchodilators.

Method—Fourteen children (6 girls, and 8 boys) who had undergone respiratory function testing in infancy were retested at school age (7–12 years). Measurements included lung volume, airways resistance, peak flow, and spirometry. Clinical problems were investigated by questionnaire. Twelve children had repeat measurements after taking salbutamol.

Results—Predominant complaints were non-productive cough and dysphagia, but even those children with major problems in infancy reported few restrictions at school or in sport or social activities. Respiratory function and clinical findings at school age appeared unrelated to status in infancy, such that even the patients with severe tracheomalacia requiring aortopexy did not have lung function testing suggestive of malacia at school age. Most patients showed a restrictive pattern of lung volume which would appear to result from reduced lung growth after surgery rather than being a concomitant feature of the primary congenital abnormality. Although six children reported wheeze and four had a diagnosis of asthma, only one responded to salbutamol. This suggests that a tendency to attribute all lower respiratory symptoms to asthma may have led to an overdiagnosis of this condition in this patient group.

Conclusion—Respiratory function testing in infancy is of limited value in medium term prognosis, but may aid management of contemporary clinical signs. In children respiratory function testing is valuable in assessing suspected asthma and effects of bronchodilators.

(Arch Dis Child 1999;81:404–408)

Keywords: thoracic gas volume; airways resistance; spirometry; cough

this condition whose birth weight was greater than 1500 g.² There is, however, associated morbidity and difficulties in management arise owing to unpredictable airway mechanics and lung growth following neonatal repair of OA-TOF. These children may develop cough, wheeze, aspiration pneumonia, vomiting, choking, feeding difficulties, and near death episodes which are most pronounced in the first 3 years of life and improve considerably after 8 years of age.³ Variable respiratory and gastrointestinal symptoms can continue into later childhood and adult life, and may be associated with abnormal pulmonary function tests.^{4–6}

There have been few investigations of respiratory mechanics in this condition in infancy. From a small number of studies, abnormalities of lung volumes, airways resistance, and flow-volume curves which correlate with clinical status have been found.⁷ Little is known about growth of the lungs and airways in this condition as, even in adulthood, there appear to be variable obstructive and restrictive defects on pulmonary function testing.⁸ The prognostic value of infant respiratory function tests in this group is unknown. The primary aim of this study was, therefore, to determine if any relation exists between respiratory function in infancy and at school age, and thus determine the value of tests performed in infancy.

Some studies have speculated on airways hyperreactivity^{8–11} but little information is available on the response to a bronchodilator in survivors of this condition. Our secondary aim was to investigate response to β_2 sympathomimetic medication in this group of children.

Patients and methods

PATIENTS

The patients in this study had their OA-TOF repair performed in Leicester. Measurements of lung mechanics were performed in 16 subjects by the age of 3 months.⁷ Fourteen of the original group of 16 survived to school age. At this follow up study the children (six girls, eight boys) were aged between 7 and 12 years. None of the group had scoliosis or chest deformity.

The study was performed with the approval of the local ethics committee.

STUDY DESIGN

This was an open study in which index cases were investigated and results compared with data obtained in infancy and with predicted values from the literature.

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Accepted 20 July 1999

Families of the 14 children were contacted by letter and invited to participate in the study. Case notes of all children were reviewed with particular reference to feeding difficulties, dysphagia, choking, respiratory symptoms including cough/wheeze, drug treatment, and number and nature of further surgery or oesophageal dilatations. Each child attended the respiratory laboratory for respiratory function tests. Patients who were taking regular bronchodilators were asked to omit their morning dose.

A questionnaire was completed for each patient seeking information about respiratory and gastrointestinal symptoms, as well as social and drug history. Height and weight were measured and a clinical examination of the respiratory system was performed.

Tests of respiratory function were then performed, comprising peak expiratory flow (PEF), spirometry, and whole body plethysmography to measure thoracic gas volume and airways resistance. Respiratory function tests were repeated following administration of a short acting β_2 agonist.

MEASUREMENTS OF RESPIRATORY FUNCTION

The procedures for measuring infant lung mechanics have been previously described.⁷ PEF was measured in school age children while standing, using a Wright's peak flow meter. The highest of three technically satisfactory measurements was recorded. Measurement of forced vital capacity (FVC), forced expiratory volume in one second (FEV_1), maximum expiratory flow at 50% and 25% vital capacity (MEF_{50} , MEF_{25}), maximum mid-expiratory flow, maximum inspiratory flow, and the ratio of mid-expiratory and mid-inspiratory flow ($V_{E50}:V_{I50}$) were measured using a simple screen pneumotachograph. Children were studied while sitting upright and wearing a noseclip. Signals were recorded onto a storage oscilloscope and subsequently plotted at slow speed onto a chart recorder and analysed by hand. In all cases, at least three technically satisfactory recordings were made and analysed according to the American Thoracic Society criteria.¹²

Measurements of thoracic gas volume (TGV), airways resistance (Raw), vital capacity, and residual volume were made with the child seated within a whole body plethysmograph (Jaeger) which incorporated a heated, humidified rebreathing system, thereby avoiding the need for panting. Mean values for Raw and TGV were calculated. The 12 children whose parents consented were then given 2.5 mg of nebulised salbutamol and the respiratory function tests were repeated after 15 minutes.

All measurements were compared with predicted values based on height and expressed as standard deviation (SD) scores.¹³⁻¹⁷

ANALYSIS OF RESULTS

The SD scores of TGV and Raw obtained at school age were compared with standard error (SE) scores of the same measurements made in infancy⁷ by paired *t* tests. The measurement of maximum expiratory flow at functional re-

sidual capacity (V'_{maxFRC}) has no direct equivalent in the data at school age but was obtained at a lung volume close to MEF_{50} and MEF_{25} , both of which can be used for comparison. The SD scores for MEF_{50} and MEF_{25} were therefore compared using *t* tests with the broadly similar SE score for V'_{maxFRC} obtained at first testing. Linear regression analysis was used to compare paired measurements to examine the predictive value of infant testing. Pearson's correlation coefficient was used to examine the strength of the relation between measured variables.

Flow volume curves were inspected for evidence of airway collapse.

Children were considered to have a positive response to salbutamol if they showed an increase in peak flow of at least 20%, an increase in FEV_1 of at least 15%, or both.

Results

CLINICAL FINDINGS AND RESPIRATORY FUNCTION

On the day of lung function testing all the children were symptom free. Table 1 summarises the distribution of current symptoms. Cough was present in the majority and described as unproductive. Four of the six children with wheeze were diagnosed as asthmatic and were on bronchodilator treatment which was used on an "as required" basis. Dysphagia occurred predominantly with bread or eating too fast. Patient 11 had more than three respiratory infections in one year and was known to have a recurrent consolidated single lung lobe. Eight children had repeated oesophageal dilatation in preschool years. Three children had repeat surgery to their TOF, one of whom (patient 12) had three operations to repair a recurrent TOF and subsequently underwent oesophageal replacement surgery with a colonic conduit. Two patients (13 and 14) had tracheopexy performed for life threatening tracheal collapse. However, even those children with many complications in infancy reported few restrictions at school or in sport, with minimal or no school loss or effect on social activities.

Results of baseline spirometry and plethysmography are shown in table 2. Patients 1 and 7 could not complete full forced expiration, and plethysmographic data could not be obtained for patient 3. Two other patients (4 and 12) completed expiration within one second so FEV_1 could not be calculated. Mean values for the group as a whole showed a

Table 1 Distribution of current symptoms at school age

Patient	Cough	Wheeze	Choking	Dysphagia	Recurrent respiratory infections
1	+	-	-	-	-
2	-	-	-	+	-
3	+	+	-	+	-
4	+	+	+	+	-
5	+	-	+	+	-
6	+	-	+	+	-
7	-	-	-	-	-
8	+	+	+	+	-
9	+	-	-	+	-
10	+	+	+	+	-
11	+	-	+	+	+
12	+	-	-	+	-
13	+	+	+	+	-
14	+	+	-	-	-

+ present; - absent.

Table 2 Baseline childhood respiratory function expressed as standard deviation scores

Patient	FVC	FEV ₁	MEF ₅₀	MEF ₂₅	TGV	TLC	Raw	PEF
1		-4.05			+0.08	-1.78	-1.32	-2.21
2	-1.02	+0.16	+0.12	+0.98	-1.46	-1.39	-1.85	-0.54
3	-1.70		+0.28	+1.97				-0.64
4	-1.53		+0.78	+1.02	+3.13	+2.41	-0.62	-0.71
5	-1.58	-0.41	-0.46	-0.06	-1.15	-0.11	-2.30	-1.50
6	-3.87	-3.66	-3.75	-2.49	-4.78	-3.11	+0.86	-2.12
7					-2.91	-1.86	-1.47	-1.41
8	-3.10	-3.08	-3.17	-2.13	-2.86	-2.99	+0.27	-2.34
9	-3.16	-2.21	-0.23	-1.47	-1.86	-3.95	-0.55	+0.03
10	-3.77	-3.04	-1.34	-1.54	-2.74	-0.79	+0.40	-1.03
11	-3.73	-2.43	-1.00	-0.13	-0.19	-1.47	-0.42	-3.14
12	-6.16		-2.45	-1.74	-0.99	-2.38	+1.16	-3.34
13	-2.93	-2.27	-1.94	-0.99	-1.85	-2.23	-0.54	-1.36
14	-2.15	-1.81	-1.33	-2.35	-2.45	-2.78	+0.90	+0.34
Mean	-3.57	-2.04	-1.64	-1.48	-2.18	-1.73	-0.98	-1.69

FVC, forced vital capacity; FEV₁, forced expired volume in 1 second; MEF₅₀ and MEF₂₅, maximum expiratory flow at 50% and 25% vital capacity; TGV, thoracic gas volume; TLC, total lung capacity; Raw, airways resistance; PEF, peak expiratory flow.

reduction in TGV, FVC, and FEV₁ which exceeded 2 SD below prediction. As a group, there appeared to be no clinical correlation with severity of symptoms in preschool years. Seven of the 10 children in whom FEV₁ could be measured had a low value, and of these three had occasional wheeze and five suffered with choking and dysphagia. Five patients had a low PEF, four of whom also had a low FEV₁ and suffered from wheezing.

None of the children had increased airways resistance. There were no abnormalities of maximum inspiratory flows and V_E:V_I ratios were normal.

Special mention should be made of patients 13 and 14 who had tracheopexy. Patient 13 had a low FEV₁ and FVC but other measurements were within normal limits. Patient 14 had a low TGV and FVC, with evidence of reduction in flow rate at low lung volume as demonstrated by a reduced MEF₂₅.

RELATION BETWEEN FINDINGS IN INFANCY AND CHILDHOOD

Table 3 summarises the respiratory function of our group of patients in infancy as well as the major clinical problems encountered in these children early in life. Even those children who had multiple and severe respiratory symptoms in infancy now have minimal restrictions on school attendance, sport, and travel. In the survivors clinical status was generally good and appeared unrelated to the severity and extent of problems in infancy, other than the observation

Table 3 Results of infant respiratory function (expressed as standard errors of prediction) with problems reported in infancy

Patient	TGV	Raw	V' _{maxFRC}	Clinical problems
1	-0.9		+0.12	
2	-1.74		+1.10	
3	-0.48	-0.36	+0.80	
4	+0.31	-2.52	-0.32	
5	+1.29	-4.00	+0.24	
6	-0.28	-0.59		Harsh cough
7	-0.03	+0.08		Noisy breathing
8	-0.52		+0.66	GOR, recurrent aspiration
9	-1.43	+1.42	-1.29	GOR, cough with feeds
10	+0.43	+5.30		GOR, aspiration pneumonia
11	+2.90	+0.04		GOR, aspiration pneumonia
12	+0.53	+1.47	-0.89	Subglottic stenosis
13	-0.18	+1.47		GOR, aspiration pneumonia, tracheomalacia
14	+2.16	+3.32		Tracheomalacia, recurrent aspiration
Mean	+0.15	+0.51	+0.05	

TGV, thoracic gas volume; Raw, airways resistance; V'_{maxFRC}, maximum expiratory flow at functional residual capacity; GOR, gastro-oesophageal reflux.

Figure 1 Regression of thoracic gas volume (TGV) score in childhood against that in infancy. Correlation (r^2) = 0.14, p = 0.65.

that six of the seven children with low FEV₁ had gastro-oesophageal reflux in infancy and/or suffered from choking spells. However, there were two infants in the original study who later died, one of whom had generalised severe tracheobronchomalacia and the poorest respiratory function in the group. This child did not survive despite undergoing tracheopexy and aortopexy. The second died following sudden collapse caused by aspiration of vomit during an episode of gastroenteritis.

When the SD scores of respiratory function indices obtained at school age were compared with SE scores of the same measurements made in infancy no differences were found for Raw or for MEF₅₀ and MEF₂₅ when compared with the SE score for V'_{maxFRC}. Regression analysis of paired measurements failed to show any relation between the values obtained in infancy and at school age. When comparing TGV measured at both ages we failed to find any correlation between the paired measurements (fig 1). However, there was a significant reduction in the mean TGV score from +0.15 in infancy to -2.18 in childhood (p = 0.013).

All children had flow-volume loops which were normal in shape, with no change following salbutamol administration. None of them showed evidence of the gross airway collapse that was present in six cases in infancy.

BRONCHODILATOR RESPONSIVENESS

Four of the six children with wheeze had a diagnosis of asthma and had been prescribed bronchodilator treatment. With the exception of one child, who had used a salbutamol inhaler six hours before attending the laboratory, none of the children had taken bronchodilators in the 24 hours preceding their visit. However, of the 12 children tested only one (patient 4) showed a response to salbutamol with a 20% increase in PEF. This child was previously diagnosed as having asthma. He was the only child with a significant increase in TGV and total lung capacity, suggesting hyperinflation. Although bronchodilators had been prescribed he was not taking treatment on a regular basis.

Discussion

The respiratory symptoms we observed such as cough, wheeze, and choking are common after OA-TOF repair and have been previously described.³⁻⁶ These may be multifactorial in origin and relate to recurrent inhalation, defective oesophageal motility, bronchial

obstruction (not necessarily asthma), and tracheomalacia. We found a lower incidence of recurrent bronchitis than previously reported.³ Although only one child (patient 7) was entirely symptom free on questioning, none of the children had major restrictions on lifestyle, and sporting and school activities. This finding applies even to those patients who had major abnormalities of lung function and repeated medical and surgical interventions in infancy.

This is the first longitudinal study to report respiratory function tests in infancy and childhood following OA-TOF repair. The abnormalities of respiratory function found in the present study are similar to those in previous studies, confirming a restrictive pattern with a decrease in lung volumes in children following OA-TOF repair.⁸⁻¹¹ The reduction in FVC is proportionally greater than that in FEV₁, again indicating a restrictive pattern.

In infancy, nine of 16 infants investigated up to three months following OA-TOF repair had abnormalities of lung function which broadly corresponded with occurrence of symptoms at that time. The symptoms were predominantly respiratory or gastro-oesophageal. Lung function abnormalities included an increase in TGV (19%), increase in Raw (38%), and abnormalities in patterns of Raw and pressure-flow loops (50%). When compared with the infant data, present findings show a weak relation between respiratory function and clinical symptoms. The occurrence of troublesome reflux in early childhood appears to be associated with a significant reduction in FEV₁ at school age, but the number of patients in the study group precludes meaningful statistical analysis.

The use of a scoring system enabled us to compare directly measurements made at both ages. The scoring system for infant data was based on standard errors of prediction¹⁸ whereas that for data at school age was an SD score. A score based on standard errors of prediction takes account of the variability in the estimated trend line as well as variability in the population. However, unless the sample size of the reference population is small, or the experimental data require extrapolation from the reference population, the two systems give very similar scores, and direct comparison between the measurements at two ages was therefore possible.

In contrast to findings in infancy,⁷ mean TGV falls below 2 SDs of the predicted value, with five individuals having a reduced value. This restrictive pattern has been previously described but the present study is the first to document a change in TGV between infancy and school age. These findings suggest that the infants are born with a normal lung volume which then falls below normal for length, presumably because of abnormal lung growth. Alternatively, the measurements made in infancy may have appeared normal because the infants were actually hyperinflated at the time of testing, which was usually within a month of primary repair. If the latter explanation was correct, it would be expected that infants who had repeat testing within the first year of life

would have a lower TGV score at one year than at first test. This was not observed,⁷ which suggests that the restrictive pattern seen at school age is the result of suboptimal lung growth in the early years of life rather than a concomitant feature of the primary congenital abnormality.

Few studies have compared measurements made in infancy with those in childhood. In a study of children with cystic fibrosis¹⁹ there was no relation between airways resistance or indices derived from forced expiration at the two ages. However, there was a significant relation between TGV measured in infancy and at school age. In contrast, a similar study of very low birth weight infants²⁰ failed to find any relation between TGV measured at the two ages. These apparent discrepancies probably arise from the different populations under study and the predicted patterns of change. Infants with cystic fibrosis may have normal respiratory function initially but then go on to show a gradual decline. Infants of very low birth weight or those with repair of OA-TOF may show early abnormalities but would not necessarily be expected to deteriorate.

In our original study group three of the six infants with stridor had patterns of airway resistance characteristic of intrathoracic upper airway obstruction, with irregular expiratory and inspiratory pattern, compatible with tracheomalacia.⁷ These three infants underwent tracheopexy for tracheomalacia. Both tracheopexy and aortopexy are successful operations in severe tracheomalacia.²¹⁻²³ One infant with additional extensive bronchomalacia went on to have an aortopexy but did not improve following the operation and subsequently died. Lung function of the two surviving children at school age showed that airways resistance was normal although TGV and FEV₁ were marginally low. Their flow-volume loops showed no evidence of airways collapse. With objective testing it appears that the obstructive irregular pattern of the infant flow-volume loops in tracheomalacia disappears with time. This may be attributable, in the short term, to tracheopexy, but is likely in the longer term to be caused by natural improvement of the airway. Cartilage development with age may contribute to increasing airway stabilisation.

Previous reports involving studies of bronchial hyperresponsiveness by methacholine challenge, histamine challenge, and β agonist administration have not found any significant correlation between pulmonary function abnormalities and/or clinical symptoms and the presence of a positive challenge in OA-TOF subjects.⁸⁻¹¹ Although we did not observe any paradoxical effect of salbutamol, even those children who were diagnosed as asthmatic had only minor improvements in lung function following salbutamol administration. A tendency to attribute all lower respiratory symptoms to asthma has probably led to an overdiagnosis of this condition within this group of patients. We emphasise the need for objective lung function tests if possible, before prescribing a bronchodilator, as not all children with cough and wheeze will have reversible airway narrowing

and bronchodilator treatment might worsen symptoms or airway function in certain children. Knowing that these children with OA-TOF repair have structurally abnormal airways means that it is essential to examine function before commencing bronchodilator treatment.

In conclusion, primary repair of OA-TOF is surgically successful. Even those children with many complications in infancy appear to have a good quality of life with few restrictions in sport, minimal (if any) school loss, or effect on social activities. Although functional abnormalities of the oesophagus and trachea persist,²⁴ these may not cause any symptoms, whereas in infancy they may cause life threatening illnesses.

Infant lung function testing may aid the assessment of contemporary clinical signs and early effects of management such as antireflux treatment. However, use of observations and testing in infancy for medium term prediction of both clinical and laboratory based respiratory dysfunction is not reliable. Long term respiratory follow up should be included in the review programme for children following OA-TOF repair in parallel with surgical gastrointestinal and general paediatric care, but where respiratory function testing is indicated it should be reassessed in preference to reliance on early measurements. Formal respiratory function testing is valuable in assessing suspected asthma and effects of bronchodilator treatment in these children.

The authors thank Mr Spearing for financial support, Mr Sumit Agrawal and Mr Ramesh Gupta for their statistical input, and Mr Michael Johnstone for allowing us to study his patients. Special thanks go to the parents and children involved in the study, for their cooperation.

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