6. Chasen ST, Al-Kouatly HB, Ballabh P, Skupski DW, Chervenak FA. Outcomes of dichorionic triplet pregnancies. ***Am J Obstet Gynecol*** 2002; 186: 765-767.
7. Howard RJ, Tuck SM, Pearson TC. Pregnancy in sickle cell disease in the UK: results of a multicentre survey of the effect of prophylactic blood transfusion on maternal and fetal outcome. ***Br J Obstet Gynaecol*** 1995; 102: 947-951.
8. Koshy M, Burd L, Wallace D, Moawad A, Baron J. Prophylactic red-cell transfusions in pregnant patients with sickle cell disease. A randomized cooperative study. ***N Engl J Med*** 1998; 319: 1447-1452.

Bronchiectasis following repair of esophageal atresia and tracheo-esophageal fistula

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The incidence of esophageal atresia and tracheoesophageal fistula (EA/TEF) was reported to be 1 in 4000-5000 live births.[1] Recurrent aspiration pneumonia is the most common complication described according to different mechanisms due to spill over of secretions through TEF or esophageal pouch, esophageal dysmotility, gastro-esophageal reflux (GER) and absence of ciliated epithelium in the trachea.[2] Many other causes of respiratory complications including esophageal dysmotility, tracheomalacia, anastomotic stricture, and recurrent or double fistula were described previously.[3,4] In this report, we describe bronchiectasis as a complication following repair of EA and TEF. We undertook a retrospective review of the charts for all EA/TEF patients referred to the pulmonary clinic for evaluation of recurrent chest infection and preoperative evaluation during the period from November 1993 to October 2004 at the main tertiary care center for referral of complicated cases in Saudi Arabia. Bronchiectasis was diagnosed based on dilated bronchi on CT chest.

There were a total of 41 patients, 26 (63%) males and 15 (37%) females. Fourty patients (98%) are alive and one (2%) died. Fourteen (34%) were premature and 27(66%) were full term. The TEF was diagnosed at birth in 34 (83%) of the patients. Patients were referred to King Faisal Specialist Hospital and Research Center at 15±29 months. The period of follow up was 5±3.8 years. Diagnosis of TEF was based on nasogastric tube coiling (NGT) and by dilated blind esophagus on chest x-ray in 40 (98%) of the patients. An EA and distal TEF were found in 37 (90%) of the patients, isolated EA in 2 (5%) and H-type fistula in 2 (5%) of the patients. Congenital anomalies were associated in 28 (68%) of the patients. Cardiac anomalies were found in 11 (27%), gastrointestinal (GIT) in 8 (20%), respiratory system anomalies in 12 (30%), renal in 7 (17%), skeletal in 12 (30%), and chromosomal in 7 (17%). More than 1/3 of the patients had post-operative complications including pneumothorax, recurrent fistula, leakage at operation site and empyema. Thirty (73%) presented with pneumonia and required prolonged ventilation. Esophageal dysmotility and GER developed in >90% of the patients. Twenty-four (60%) of the patients required Nissen fundal plication for GER. Esophageal stricture that required >3 dilatations developed in 16 (46%) of the patients. The GER was significantly related to development of atelectasis, dysmotility, and aspiration pneumonia ($p<0.05$), but not related to surgery type if it is primary anastomosis or staged surgery ($p>0.05$). Pulmonary complications developed in >70% of the patients including persistent atelectasis, chronic aspiration pneumonia, asthma or hyper reactive airway disease, and chronic lung disease that required oxygen for more than one month. Tracheomalacia occurred in 12 (29%) of the patients. Bronchiectasis developed in 7 (17%) of the patients (**Table 1**), 2 of them after gastric tube replacement of esophagus, one after colonic replacement, and 4 developed after primary repair. Two of the 4 patients with primary repair were premature, another one with multiple congenital anomalies and the 4th one with recurrent fistula, esophageal diverticulum and cardiac anomalies (**Table 1**). Pulmonary function test (PFT) was carried out in 16 (40%) patients who were able to comprehend the test maneuver. Eighty-eight percent of patients who performed PFT showed abnormal values: obstructive PFT changes in 3 (7%), restrictive in 8 (20%), combined obstructive and restrictive changes in 3 (7%) and normal in 2(4%).

Long-term pulmonary complications have been described before.[2,4-6] Couriel et al[4] described bronchitis for more than 8 years in 5/20 patients (25%), and denoted that lung disease improves with time. Chetcutti et al[5] described asthma development in 40/155 (26%) patients after TEF repair, with restrictive lung changes in 18 (12%) of the population. Delius et al[2] showed that 31/68 patients (46%) developed recurrent pneumonia that required 1-10 admissions to hospital for treatment. Robertson et al[6] performed PFT in 25 patients with TEF repair and their siblings and found that, although PFT values were within normal limits, they were significantly different compared to their siblings. The later study also showed that 6/25 patients had positive methacholine challenge test as a sign of obstructive airway disease and 9/25 had a restrictive pattern. Gastroesophageal reflux as the primary cause of respiratory symptoms in these patients has

Table 1 - Bronchiectasis after tracheoesophageal fistula repair.

Type of anastomosis	Lobes involved	Comments
Gastric tube	RLL, RML, LLL	Operated in a local hospital, long gap EA, leakage at operation site, stricture resection, septicemia
Gastric tube	Left lung bronchiectasis, RML atelectasis	Left lung pneumonectomy due to bronchiectasis, recurrent aspiration, GER, kidney abnormalities with repeated UTI, stricture of gastric tube, seizure disorder, tracheomalacia
Colonic placement	LUL, RUL	VATER association, VUR grade IV left kidney, left nephrectomy,
Primary	RUL	E-diverticulum, recurrent fistula, GER, E -stricture, needed 11 dilatations, VSD closure
Primary	RML, LLL, RLL	Premature 34 weeks, polyhydramnios, operated in a local hospital, hiatal hernia, asthma, GER
Primary	RML, LLL	Congenital anomalies, choanal atresia, tracheomalacia and tracheal stenosis, tracheal stent, GER, fundal plication, microcephaly, developmental delay
Primary	LLL	Premature 34 weeks, RDS, RSV bronchiolitis, coagulase positive *Staphylococcus aureus* septicemia, GER.

E - esophageal, GER - gastroesophageal reflux, RDS - respiratory distress syndrome, VSD - ventricular septal defect, VUR - vesicoureteric reflux, RSV- respiratory syncytial virus, RML - right middle lobe, RUL - right upper lobe, LLL - left lower lobe, RLL - right lower lobe, VATER - vertebral, anal, tracheal, esophageal, renal and radial association

been implicated in some reports.[4,7] Tracheomalacia and recurrent fistula are known to occur in these patients as a cause of respiratory complications,[2] however, the incidence and relative significance of these multiple causes of respiratory symptoms are unclear.

Our report has shown that bronchiectasis post TEF repair developed in 7 (17%) of the patients. It requires long-life follow-up and antibiotic prophylaxis. This could be explained in view of recurrent infections, persistent atelectasis, and recurrent aspirations due to GER that masked the early recognition of such complication. Prematurity and a complicated clinical course have contributed significantly in the cause of bronchiectasis in our population. Another important cause is the replacement of the esophagus with gastric tube or colon, which needs to be addressed again as the last option in staged surgery unless spontaneous elongation of esophagus fails. Such patients need to be followed for an undetermined period to ensure that such complications are avoided and treated early.

In summary, bronchiectasis is a common complication after TEF repair and is associated with significant morbidities that need to be recognized and managed early before significant and irreversible damage develops.

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References

1. Spitz L. Esophageal atresia: Past, present, and future. ***J Pediatr Surg*** 1996; 31: 19-25.
2. Delius R, Wheatley M, Coran A. Etiology and management of respiratory complications after repair of esophageal atresia with tracheoesophageal fistula. ***Surgery*** 1992; 112: 527-532.
3. Engum SA, Grosfeld JL, West KW, Rescorla FJ, Scherer LR 3d. Analysis of morbidity and mortality in 227 cases of esophageal atresia and/or tracheoesophageal fistula over 2 decades. ***Arch Surg*** 1995; 130: 502-508.
4. Couriel J, Hibbert M, Olinsky A, Phelan P. Long term pulmonary consequences of oesophageal atresia with tracheo-oesophageal fistula. ***Acta Paediatr Scand*** 1982; 71: 973-978.
5. Chetcutti P, Phelan P, Greenwood R. Lung function abnormalities in repaired oesophageal atresia and tracheoesophageal fistula. ***Thorax*** 1992; 47: 1030-1034.
6. Robertson D, Mobaireek K, Davis, GM, Coates A. Late pulmonary function following repair of tracheoesophageal fistula or esophageal atresia. ***Pediatr Pulmonol*** 1995; 20: 21-26.
7. Whitington PF, Shermeta DW, Seto DSY, Jones L, Hendrix TR. Role of lower esophageal sphincter incompetence in recurrent pneumonia after repair of esophageal atresia. ***J Pediatr*** 1977; 91: 550-554.