Esophageal foreign bodies in Yemen

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Foreign body (FB) impaction in the esophagus is a common and serious health problem. Foreign bodies can lead to many complications such as esophageal perforation, mucosal erosions and fistulizations. Complication from esophageal FB can include death. The maximum incidence of esophageal FB occurs among children under 5 years old. The most common kind of the esophageal FBs in Oman are mutton bones (29.5%), fish bones (20.5%), and coins (20.5%). In Saudi Arabia, these are fish bones (27%), meat bones (23%), and coins (12%). In Jordan, coins (55.5%), bones (13.9%), and metallic objects (6.7%). The general objective of this study is to describe the distribution of the esophageal FB among patients presenting at Al-Thawra Teaching Hospital. Specifically, to classify the kinds of common FB in the esophagus, identify duration, clinical presentation and diagnosis, outcome of the interventions and to correlate the incidence of FBs with age and gender.

A total of 224 patients with FB in the esophagus attended the General Emergency Room and ENT clinic at Al-Thawra Teaching Hospital. Cases were collected prospectively from January 1st 2003, to August 31st 2004. Routine cervical and chest x-ray were obtained in most of the cases. For patients with negative x-ray, further radiographic investigations were performed (barium swallow and CT scan). All patients with a history of esophageal FB were considered emergencies, and interventions were performed in the operating theater. The vast majority of the coins were removed under direct laryngoscopic vision and McGill forceps, maintenance anesthesia was accomplished with 2-3% halothane and 100% oxygen inhalation in the operating room. For other patients, the FBs were removed by rigid esophagoscopy under general anesthesia. Patients with postoperative complications like esophageal perforation and tracheo-esophageal fistula were referred to the general surgery department. Control x-rays were carried out in some cases. The most common age group was under 5 years old, accounting for 68.6% of the cases and the youngest patients were 3 months old, while the oldest one was 70 years old. There were 120 males (53.6%) and 104 females (46.4%). As for duration, 128 (57.1%) of the patients reached the hospital on the same day of FB swallowing, while 35 (15.6%) do not know about the presence of the FB and presented with another complaints and were discovered accidentally. Some (47 [21.1%]) patients arrived between the 2nd and 4th day of FB ingestion, and only 14 (6.3%) patients came after more than 5 days. Regarding symptoms, the most common presenting symptoms for patients with esophageal FB were dysphagia 180 (80.4%), followed by odynophagia 132 (58.9%) then hypersalivation 72 (32.1%). The x-ray findings were positive in 184 (82.1%) cases, barium swallow was performed in 2 (0.9%), and CT scan in 2 (0.9%). The vast majority of FBs, 178 (79.5%), were located in the upper esophagus below the cricopharyngeus, 30 (13.4%) in the middle esophagus, and 13 (5.8%) in the lower esophageal sphincter. We found 2 (0.9%) FBs in the stomach and could not identify the exact location in 3 (1.3%) cases. Out of 145 coins removed, 132 (91%) were under direct laryngoscopic vision and McGill forceps, maintenance anesthesia was accomplished with 2-3% halothane and 100% oxygen inhalation. The diameter of the coins were between 2.2-2.5 cm. We used rigid esophagoscopy under general anesthesia in 92 (41%) cases and were successful in 86 (93.5%) cases. Four patients (1.8%) had complications, one (0.4%) patient with esophageal perforation with mediastinitis, one (0.4%) with tracheoesophageal fistula, and 2 (0.9%) with erosions without perforation. None underwent surgical removal and no death. As for the kinds of the FBs;

<table>
<thead>
<tr>
<th>Kind</th>
<th>n (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Coins</td>
<td>145 (64.7)</td>
</tr>
<tr>
<td>Fish bone</td>
<td>16 (7.1)</td>
</tr>
<tr>
<td>Meat bolus</td>
<td>11 (4.9)</td>
</tr>
<tr>
<td>Button</td>
<td>8 (3.6)</td>
</tr>
<tr>
<td>Piece of plastic</td>
<td>7 (3.1)</td>
</tr>
<tr>
<td>Piece of metal</td>
<td>6 (2.7)</td>
</tr>
<tr>
<td>Pins</td>
<td>6 (2.7)</td>
</tr>
<tr>
<td>Safety pins</td>
<td>5 (2.2)</td>
</tr>
<tr>
<td>Nail</td>
<td>4 (1.8)</td>
</tr>
<tr>
<td>Hair holder</td>
<td>3 (1.3)</td>
</tr>
<tr>
<td>Meat bone</td>
<td>3 (1.3)</td>
</tr>
<tr>
<td>Hand watch</td>
<td>2 (0.9)</td>
</tr>
<tr>
<td>Stone</td>
<td>2 (0.9)</td>
</tr>
<tr>
<td>Denture</td>
<td>1 (0.4)</td>
</tr>
<tr>
<td>Glass beads</td>
<td>1 (0.4)</td>
</tr>
<tr>
<td>Watch battery</td>
<td>1 (0.4)</td>
</tr>
<tr>
<td>Piece of wood</td>
<td>1 (0.4)</td>
</tr>
<tr>
<td>Apricot seed</td>
<td>1 (0.4)</td>
</tr>
<tr>
<td>Orange skin</td>
<td>1 (0.4)</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>224 (100)</strong></td>
</tr>
</tbody>
</table>
145 (64.7%) were coins, 16 (7.1%) were fish bones, and 11 (4.9%) were meat bolus (Table 1). The vast majority 10 (90.9%) of the patients with meat bolus were men aged 55 years and above. The patients with safety pins 5 (2.2%) were women all below 16 years old. The highest incidence of FBs (68.6%) was found among children under 5 years old. This age group is curious and tries to interact with the surroundings. Less than a half of the patients (43%) presented at the same day of FB aspiration. The reason behind this delay could be due to unawareness about the problem, living in remote, mountainous and inaccessible areas, or could be due to seeking traditional healing solutions. In our study, 10 (4.4%) of the patients presented with dysphagia and 6 (2.7%) with cough. Therefore, esophageal FBs patients may present in respiratory distress and may be treated for bronchitis or asthma. Thus, it is important to differentiate dysphagia and cough due to respiratory pathology and esophageal FBs impaction. Asymptomatic patients with esophageal FBs were (7.1%). Some of these FBs have been long standing, these patients may have grown accustomed to the presence of the FBs, and the symptoms are related to the size, kind, and location of the FBs in the esophagus. A high percentage of coins (91%) were removed under direct laryngoscopic vision and McGill forceps without general anesthesia. In our experience, this technique is safe and effective for removing cervical esophageal coins. For other esophageal FBs, we used rigid esophagoscopy. It was successful in 93.5%, and we found that rigid esophagoscopy is safe, quick, easy, and with minimum trauma. We had only 4 (1.8%) complications due to the trial for removing sharp impacted FBs by inadequately experienced hands. Coins (145 [64.7%]) were the most common FBs in the esophagus and this finding is comparable with other studies in Jordan, while higher than the proportions reported from Saudi Arabia and Oman.

In our study 5 (2.2%) FBs were safety pins and swallowed by women. In our country, females practice the wearing of the scarf, and they hold the safety pin between their teeth or lips and, can unintentionally swallow the pin.

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References


Mycobacterium tuberculosis and CD4+ T-lymphopenia. A grave combination

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Tuberculosis (TB) is still a common health problem in many parts of the world including Saudi Arabia. According to some of the estimates approximately one third of the world’s population is infected with mycobacterium tuberculosis (MTB), with 6-8 million new active cases each year, and accounting for 2-3 million deaths each year. Infection with MTB has protean manifestation, with single or multiple organ system involvement. Hematological abnormalities with the disease have been enlightened by many studies. Correction of these abnormalities with initiation of treatment indicates a good response of the disease. In the present study, we looked at the occurrence of CD4+ T-lymphopenia with active MTB infection.

We included in this study, all patients admitted in the Department of Medicine, under care of Infectious Diseases Unit, at King Khalid University Hospital, Riyadh, Kingdom of Saudi Arabia from July 2002 to June 2003 with culture proven MTB infection, and CD4+ T-cell count <300 x 10^3/L. We tested the human immunodeficiency virus (HIV) status of all the patients by western blot method and excluded those with a positive result. In addition, the patients on immunosuppressive drugs for any reason were excluded from the study. A tuberculin skin test (TST) was carried out for all the patients.

by injecting 5 units of purified protein derivative (PPD) intradermally and read after 48 hours, an induration size of 5 mm or more was considered as positive. All the anti-TB drugs (calculated as per body weight of the patient) were given orally (PO) once daily (OD). For the initial 2 months of the treatment, all the patients received 4 drugs; namely, isoniazid (INH), rifampicin (RIF), ethambutol (ETB), and pyrazinamide (PZA) and later on, INH and RIF to complete their treatment. The CD4+ T-cell count was tested before starting anti-TB drugs and at 3 and 6 months after starting the treatment. All the patients were followed in the infectious diseases clinic for a minimum of one year. The total number of the patients admitted with active MTB infection over this time was 31, and 6 of them were found to have CD4+ T-cell count <300 x 10^6/L, along with negative HIV serology, giving an incidence of 19% for this severe CD4+ T-lymphopenia with active MTB infection. The patient information and treatment outcome is given in Table 1. The CD4+ T-cells are considered to be the primary T-cell subset responsible for regulating the immune response to MTB. The HIV pandemic provides direct evidence that loss of CD4+ T-cell number and function resulted in progressive primary infection, reactivation of endogenous MTB, and enhanced susceptibility to re-infection. Many studies in mice also have confirmed this protective role of CD4+ T-cells against MTB. However, active MTB infection can lead to CD4+ T-lymphopenia with counts as low as 300 x 10^6/L, the incidence of which has been reported by Kony et al as 14.4%, whereas, according to our results it is 19%. The mean CD4+ T-cell count in our patients at the time of presentation was 139.5 ± 59.8 x 10^6/L. The patients, that expired had a CD4+ T-cell count of <150 x 10^6/L, whereas, those who survived had CD4+ T-cell count >150 x 10^6/L. Our results further support the previous findings of Pilheu et al, showing high mortality in patients with CD4+ T-cell count <300 x 10^6/L, and severe pulmonary TB. We further suggest that, those with CD4+T-cell count <150 x 10^6/L are likely to have even higher mortality. All the patients who survived in our study started to recover their CD4+ T-cell count by 3 months, which returned to normal with in 6 months. This finding shows that the CD4+ T-lymphopenia produced by active MTB infection is reversible with the effective treatment of the disease. The mean age of the patients in our study was 37.1 years, and none of them had any co-morbid systemic disease. Although military TB is usually considered to be the disease of the elderly (or very young), and immunocompromised patients, the findings of our study suggest that, severe mycobacterial disease (pulmonary or disseminated) along with the MTB induced CD4+ T-lymphopenia is more common in relatively young patients. This might be explained by the fact that young, genetically susceptible, patients probably mount more vigorous immunological response to the infection with MTB, leading in turn to more severe depletion of antigen responsive T-cells (due to the phenomenon of compartmentalization and selective increased programmed cell death of activated T-cells), and this leads to the spread of the disease (intra- or extrapulmonary), due to the lack of production of interferon-gamma and interleukin-2 from CD4+ T-cells. These are the cytokines known for helping in granuloma formation and thus limiting spread of the disease. The mean time taken for presentation from the start of symptoms in our patients was 5.5 months. This long period of active infection before seeking medical advice also, might have contributed to the development of CD4+ T-lymphopenia in

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age in years</th>
<th>Gender</th>
<th>Time of first presentation from the start of symptoms</th>
<th>Percent body weight lost before presentation</th>
<th>TB site</th>
<th>Time taken for defervescence (in days) after starting anti-TB drugs</th>
<th>CD4+ T-cell count x 10^6/L before and after starting anti-TB drugs</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>22</td>
<td>F</td>
<td>6 months</td>
<td>26.6</td>
<td>P</td>
<td>20</td>
<td>139.5 ± 59.8</td>
<td>died</td>
</tr>
<tr>
<td>2</td>
<td>33</td>
<td>M</td>
<td>8 months</td>
<td>24.2</td>
<td>P + EP</td>
<td>60</td>
<td>552 ± 83.9</td>
<td>recovered</td>
</tr>
<tr>
<td>3</td>
<td>70</td>
<td>M</td>
<td>3 months</td>
<td>22.2</td>
<td>P + EP</td>
<td>148</td>
<td>910 ± 47.4</td>
<td>recovered</td>
</tr>
<tr>
<td>4</td>
<td>26</td>
<td>F</td>
<td>5 months</td>
<td>16.6</td>
<td>P + EP</td>
<td>180</td>
<td>952 ± 47.4</td>
<td>recovered</td>
</tr>
<tr>
<td>5</td>
<td>35</td>
<td>F</td>
<td>4 months</td>
<td>20</td>
<td>P + EP</td>
<td>151</td>
<td>893.7 ± 47.4</td>
<td>recovered</td>
</tr>
<tr>
<td>6</td>
<td>37</td>
<td>M</td>
<td>7 months</td>
<td>28.5</td>
<td>EP</td>
<td>173</td>
<td>436 ± 47.4</td>
<td>recovered</td>
</tr>
</tbody>
</table>

Mean ± SD: Patient Age in years 37.1 ± 17, Time of first presentation from the start of symptoms 5.5 ± 1.8 months, Percent body weight lost before presentation 26.6 ± 17.4, Time taken for defervescence after starting anti-TB drugs 34.2 ± 17.4, CD4+ T-cell count x 10^6/L 139.5 ± 59.8, Outcome died 552.5 ± 83.9, recovered 893.7 ± 47.4.

F - female, M - male, TB - tuberculosis, SD - standard deviation, P - pulmonary, EP - extra-pulmonary
MTB induced CD4+ T-lymphopenia


Management of difficult airway in a child with arthrogryposis multiplex congenita during general anesthesia

Remziye Sivaci, MD, Canan Balci, MD, Gokhan Maralcan, MD, Ilhami Kuru, MD.

Arthrogryposis multiplex congenita (AMC) consists of complex congenital anomalies characterized with multiple contractures. The possibility of autosomal dominant inheritance with reduced penetrance is suggested for this apparently new syndrome. This syndrome may include foot deformities such as pes equinovarus (PEV), and congenital convex pes valgus. There may be a variety of deformities of the knee: flexion deformity, genu recurvatum, and genu valgum. Hip deformities such as unilateral or bilateral dislocation contractures may also exist. Craniofacial abnormalities, multiple joint contractures, pulmonary hypoplasia, cryptorchidism, and unusual ophthalmological findings are the other characteristics of this syndrome. In some severely affected persons, the central nervous system may also be affected. Arthrogryposis multiplex congenita, which is diagnosed at birth presents with multiple joint contractures. Anesthetic management of these patients requires special care. As this disease often progresses until dysfunction of multiple organ systems occur, it may have an impact on the anesthetic management. Difficult tracheal intubation may be encountered due to limited neck extension, inadequate mouth opening, and short epiglottis.

A 3-year-old and 20 kg weight boy who was diagnosed as PEV was prepared for surgery. The possibility of autosomal dominant inheritance with reduced penetrance is suggested for this apparently new syndrome. This syndrome may include foot deformities such as pes equinovarus (PEV), and congenital convex pes valgus. There may be a variety of deformities of the knee: flexion deformity, genu recurvatum, and genu valgum. Hip deformities such as unilateral or bilateral dislocation contractures may also exist. Craniofacial abnormalities, multiple joint contractures, pulmonary hypoplasia, cryptorchidism, and unusual ophthalmological findings are the other characteristics of this syndrome. In some severely affected persons, the central nervous system may also be affected. Arthrogryposis multiplex congenita, which is diagnosed at birth presents with multiple joint contractures. Anesthetic management of these patients requires special care. As this disease often progresses until dysfunction of multiple organ systems occur, it may have an impact on the anesthetic management. Difficult tracheal intubation may be encountered due to limited neck extension, inadequate mouth opening, and short epiglottis.

A 3-year-old and 20 kg weight boy who was diagnosed as PEV was prepared for surgery.
Management of difficult airway in a child with AMC during general anesthesia

in the early period of her pregnancy. The child had limitation in range of motions of his ankle, neck, hip, and knee since the neonatal period. We determined pale skin, dimorphic appearance of face, small and firm chin, stiffness, and limitation of range of motion of cervical spine, contractures in lower extremities, and difficulties in walking in physical examination. Any pathology of respiratory and cardiovascular system has not been established. We interpreted biochemical study, cranial CT, cranial MRI, and electroencephalogram as normal. Before the operation 0.2 mg/kg midazolam diluted with serum physiologic was given by rectal route. Intravenous injection (IV) route was established with 500 cc isolex P using 22-G catheter on left hand dorsal side 30 minutes after premedication. He was monitored to perform electrocardiogram (ECG), noninvasive blood pressure (NIBP), end-tidal carbon-dioxide (EtCO₂) and temperature in operating theater were in normal levels. For induction of anesthesia 10 µg/kg atropine, 3.0-mg/kg propofol, 2.0 µg/kg fentanyl, and 0.1 mg/kg vecuronium bromure were given via IV route and ventilation of the patient was supplied with oxygen 100% using facemask. Two minutes after administration of muscle relaxant, laryngoscopy was performed for intubation, but the he could not be intubated as epiglottis and vocal cords were not visualized. Ventilation of the patient was continued, and intubation manipulation was repeated. For intubation a fiberoptic laryngoscope, which has a blade that has an extension angle of 70° was used (Flexotype, Heine, Germany). He was intubated by the help of a guide inserted into a disposible tube of 4.5 ID diameter, and connected to an automated ventilator after setting tidal volume as 8 ml/kg (Cicero EM, Drager/Germany). Anesthesia was maintained with sevoflurane 2%, and nitrous oxide 60% in oxygen. The airway pressure, respiratory rate, tidal volume, EtCO₂, and anesthetic gases were monitored and EtCO₂ was normal after intubation. The EtCO₂ at remained 40-53 mm Hg during operation. The airway pressure of the patient began to increase following intubation, after 30 minutes airway pressure exceeded 20 cm H₂O, and vecuronium bromure 0.3 mg/kg was administered in bolus. The airway pressure did not decrease although the dose of muscle relaxant was repeated every 30 minutes throughout the operation, which lasted 120 minutes. He remained stable hemodynamically during the preoperative period. At the end of the operation when he entered into the awakening period, his muscle power did not improve, and atropine 10 µg/kg, and neostigmin 20 µg/kg were given in bolus for recurarisation. He was taken to the recovery room after the values of SpO₂ were normalized. He was observed until he was hemodynamically stable.

Arthrogryposis multiplex congenita is a congenital disorder that consists of multiple orthopedic anomalies. They are the candidates for difficult intubation due to small chin, stiffness of temporomandibular joint, and limitation of range of motion of cervical spine.¹ In the literature, 10 patients have been reported to present difficulty for their airway control related to AMC. Therefore, we should prefer fiberoptic laryngoscope, with a blade that has an extension angle of 70 degrees. Vocal cords can be visualized better and with the help of this type of laryngoscope can easily make intubation. The surgical manipulation would not be comfortable due to an inadequate muscle relaxation in the surgical areas. This situation may possibly be due to out of junction receptors. We think that infusion of neuromuscular blocker agents may provide better relaxation with 0.2 mg/kg/h than repeated fractional bolus ejection at 30-minute times. Therefore, regional anesthesia can be safely used for different surgical procedures in patients with AMC in the presence of appropriate monitoring.¹ When volatile anesthetic agents are used, the incidence of malignant hyperthermia is high due to scoliosis, abnormal subcutaneous tissue, and myopathic muscles. Up to now, 4 patients with AMC was reported to have hyperthermia. Furthermore, these patients may have accompanying congenital cardiac pathology in the rate of 10%, and this condition is very important in the anesthesia.¹,² Hopkins et al, reported that 2 patients with AMC developed hypermetabolic reactions during anesthesia and surgery and they proposed that the reaction is distinct from malignant hyperthermia and independent of the anesthetic agents used.³ It is suggested in the anesthetic management with AMC, low-dose ketamine be administered by continuous infusion, with a satisfactory result due to myopathic features, hypoplastic musculature, severe scoliosis, and expected increased sensitivity to various seda-tives and volatile anesthetic agents. However, propofol can be a reliable agent for anesthesia in AMC patients who had hyperthermia and difficult intubation in previous surgical procedures.³

In conclusion, AMC, which is commonly associated with other diseases characterized with accumulation of abnormal material in the joint tissues, may cause increasingly severe airway problems with age. Anticipating and preparing for a difficult airway is probably more important than using any particular anesthetic technique. As vocal cords can be well visualized and intubation can easily be made with the help of fiberoptic laryngoscope, which has a blade that has an extension angle of 70, this type of laryngoscope should be preferred. However, it is recommended that the facilities for fiberoptic intubation and an experienced pediatric anesthetist are present for all such cases. In addition,
we think that infusion of neuromuscular blocker agents may provide better relaxation than fractional bolus ejection.

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References


Sickle cell disease in a woman with triplet pregnancy

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Triple pregnancies are by nature rare, reported as 0.1% in a large series of cases. Assisted reproductive technologies have multiplied their rate of occurrence in the last 2 decades. Whether spontaneous or induced, triplet pregnancies present with obstetrical and neonatal problems several fold more often than singleton or even twins. The rate of preterm birth among triplet pregnancies are reported as 96% and still birth rate 33.8%. The major neonatal morbidity associated with triplet gestations is preterm delivery and low birth weight. Pregnancy in patients with hemoglobinopathy is associated with increased risk of maternal and perinatal morbidities and mortalities. Multiple pregnancy is potentially more hazardous than singleton pregnancy. There is a dearth of information concerning multiple pregnancies in patients with hemoglobinopathy.

We present a case of spontaneous triplet pregnancy in a patient with sickle cell disease. A 26-year-old Omani lady, gravida-2, para-1 was referred to the Obstetrics outpatient clinic of Sultan Qaboos University Hospital, Sultanate of Oman at 14 weeks of gestation. She has a known case of sickle cell disease and an ultra sonogram in a peripheral hospital at 9 weeks showed a triplet pregnancy. Her booking hemoglobin was 6.9 gm/dl and she received 2 units of packed red blood cell (PRBC) transfusion in the referring hospital. Her menstrual cycles were regular and she has not used any ovulation inducing agents or contraceptives.

Her first pregnancy was also spontaneous, with a term delivery of a male baby weighing 2700 gm a year ago. She had postpartum hemorrhage requiring 2 units of PRBC. She had several admissions during that pregnancy for vaso-occlusive crisis or for blood transfusions. The patient was diagnosed to have sickle cell disease S/βthal since childhood, requiring several admissions to the hospital. Her 4 siblings are also suffering from the same disease. She had a non-consanguineous marriage and her husband is normal. Clinical examination revealed pallor and a uterine size corresponding to 24 weeks of gestation. Ultrasound examination showed 3 viable fetuses of 14 weeks gestation and a prophylactic cervical cerclage was inserted after a week. She continued to have regular follow up with hematologist and obstetrician and was put on oral penicillin V and folic acid supplements. Serial ultrasonogram showed satisfactory growth of all 3 fetuses. She required several admissions for top up and exchange transfusions. There was no evidence of any infection during pregnancy. At 26 weeks she was given 2 doses of injection dexamethasone to promote fetal lung maturity in case she goes into preterm labor. An elective cesarean section was planned at 32-34 weeks of gestation, but at 30 weeks she was admitted with labor pains. Cervical cerclage was removed and an emergency lower segment cesarean section was performed after arranging full neonatal support.

The details of the newborn babies are shown in Table 1. The placenta was trichorionic triamniotic weighing 1100gms. The estimated blood loss was 600 mls and she received 2 units of PRBC transfusion after surgery. Received 300 µgm of anti-D as she was Rh negative and without antibodies. Postoperative period was uneventful and she was discharged on the 6th day. At 6 weeks postpartum checkup she was clinically well, lactating and started on injection Depo-Provera for contraception. Babies at 3 months of age showed normal serial growth in all parameters and there was no evidence
of any retinopathy of prematurity. The incidence of multi-fetal gestation has increased significantly since the introduction of ovulation induction therapy and in-vitro fertilization and embryo transfer techniques. Multi-fetal gestation is associated with increased frequency of maternal complications and higher perinatal morbidity and mortality. As the number of fetus increases, the duration of gestation and birthweight decreases. The mean gestation at birth for triplets was reported as 33.5 ± 2.3 weeks and the mean birth weight as 1810 ± 270 gm for monochorionic pairs compared to 2125 ± 265 gm for dichorionic triplets. Our triplets were born earlier at 30 weeks and the mean birth weight was lower, only 1323 gm probably due to the sickle cell disease. Pregnancies complicated by sickle cell disease were significantly more likely to be associated with anemia, preterm delivery, proteinuric hypertension, birth weight below the 10th centile and emergency cesarean section. Severe sickling complications occurred more commonly in third trimester and there was some evidence that a prophylactic transfusion program reduced this risk. However, prophylactic transfusion did not improve obstetric outcome when compared with those pregnancies that were not transfused. Koshy et al did not find any significant difference in perinatal outcome between the offspring’s of mothers with sickle cell disease who received prophylactic transfusion compared to those who did not receive. Prophylactic transfusion significantly reduced the incidence of painful crises of sickle cell disease but the increase in cost, number of hospitalizations and the risk of alloimmunization were disadvantages of prophylactic transfusion. Our patient was admitted several times with crises or anemia and had several exchange and simple transfusions during pregnancy. We do not follow a policy of routine prophylactic transfusion but given for selected cases with previous poor obstetric outcome.

Meticulous care by obstetrician and hematologist with increased fetal surveillance and elective cesarean delivery are suggested in the management of such patients. Also, with the neonatal complications documented in these preterm babies, a highly functional neonatal intensive care unit is necessary.

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References


Table 1 - Details of the triplets.

<table>
<thead>
<tr>
<th>Presentation</th>
<th>Gender</th>
<th>Weight</th>
<th>Apgar</th>
<th>Problems</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cephalic</td>
<td>Girl</td>
<td>1400 gm</td>
<td>7 at 1 minute - 8 at 5 minute</td>
<td>RDS, metabolic acidosis. Ventilated x 15 hr.</td>
<td>Discharged on 35th day. Weight 1645 gm. At 40 weeks corrected age weight 2950 gm, height 45 cm.</td>
</tr>
<tr>
<td>Cephalic</td>
<td>Boy</td>
<td>1270 gm</td>
<td>7 at 1 minute - 8 at 5 minute</td>
<td>RDS, metabolic acidosis. Ventilated x 15 hr.</td>
<td>Discharged on 35th day. Weight 1820 gm. At 40 weeks corrected age weight 3180 gm, height 49 cm.</td>
</tr>
<tr>
<td>Cephalic</td>
<td>Girl</td>
<td>1300 gm</td>
<td>7 at 1 minute - 8 at 5 minute</td>
<td>RDS, metabolic acidosis. Ventilated x 15 hr.</td>
<td>Discharged on 35th day. Weight 1830 gm. At 40 weeks corrected age weight 3080 gm, height 48 cm.</td>
</tr>
</tbody>
</table>

RDS - respiratory distress syndrome.
Congenital anomalies were associated in 28 (68%) and H-type fistula in 2 (5%) of the patients. Bronchiectasis was found in 37 (90%) of the patients, isolated EA in 2 (9%) of the patients. An EA and distal TEF were based on dilated blind esophagus on chest x-ray in 40 patients (98%). TEF was based on nasogastric tube coiling (NGT) in 27 (66%) of the patients. Bronchiectasis developed in 7 (17%) of the patients. 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 in 25 patients with TEF repair 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. Couriel et al described bronchitis for more than 8 years in 5/20 patients (25%), and denoted that lung disease improves with time. Chetcutti et al described asthma development in 40/155 (26%) patients after TEF repair, with restrictive lung changes in 18 (12%) of the population. Delius et al showed that 31/68 patients (46%) developed recurrent pneumonia that required 1-10 admissions to hospital for treatment. Robertson et al 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 been described in 4/5000-5000 live births. 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. Many other causes of respiratory complications including esophageal dysmotility, tracheomalacia, anastomotic stricture, and recurrent or double fistula were described previously. 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. Forty 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%).

The incidence of esophageal atresia and tracheoesophageal fistula (EA/TEF) was reported to be 1 in 4000-5000 live births. 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. Many other causes of respiratory complications including esophageal dysmotility, tracheomalacia, anastomotic stricture, and recurrent or double fistula were described previously. 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.

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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.
been implicated in some reports.\textsuperscript{4,7} Tracheomalacia and recurrent fistula are known to occur in these patients as a cause of respiratory complications,\textsuperscript{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


\begin{table}
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\begin{tabular}{|llp{10cm}|}
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Type of anastomosis & Lobes involved & Comments \\
\hline
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 UTH, 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, polyhydramsinos, 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 \textit{Staphylococcus aureus} septicemia, GER. \\
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\caption{Bronchiectasis after tracheoesophageal fistula repair.}
\end{table}

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