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Spontaneous pneumothorax in a Neonate with TEF and Double Oesophageal Atresia. A Case Report

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Symptomatic Spontaneous pneumothorax is uncommon disease scenario in term neonates. But pneumothorax can occur in neonates who had CPR or are on mechanical ventilator for different causes of respiratory distress. Congenital double esophageal atresia is a very rare type of esophageal atresia. If only single membranous atresia it can be isolated esophageal membranous atresia or with trachea-esophageal fistula. This type of atresia was not mentioned in gross or Vogt classification. Previously, only a few cases with an obstructing web had been reported and also multiple esophageal atresias. But there is no any documented case report with TEF & multiple esophageal atresia. The choices of treatment vary from ligation of the fistula with excision of the membrane by esophagotomy and/or gastrotomy, thoraco-abdominal approach and end to end anastomoses of the proximal & distal atresia. Here we present a case of a neonate who had developed respiratory distress since birth with a cause of tension spontaneous pneumothorax and managed by a chest tube and also diagnosed to have oesophageal atresia with distal tracheo-esophageal fistula on further evaluation and showing additional distal membranous oesophageal atresia during the thoracotomy. And he was managed accordingly.

Key words: Spontaneous Pneumothorax, esophageal atresia, Membranous atresia, TEF

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Introduction

Symptomatic Spontaneous pneumothorax is uncommon disease scenario in term neonates. But pneumothorax can occur in neonates who had CPR or are on mechanical ventilator for different causes of respiratory distress [1,2].

Congenital anomalies of the esophagus occur in approximately 1 in 3500 live births, with some geographical variance 3. The most common type (84%) is an esophageal atresia (EA) with a distal tracheoesophageal fistula (TEF).Congenital double esophageal atresia is a very rare type of esophageal atresia.If only single membranous atresia it can be isolated esophageal membranous atresia or with tracheoesophageal fistula. But this type of atresia was not mentioned in gross or Vogt classification. Previously, only a few cases with an obstructing web had been reported[4,5,6] and also multiple esophageal atresia[7]. But there is no any documented case report with TEF & multiple esophageal atresia. The choice of treatment varies from ligation of the fistula with excision of the membrane by esophagotomy and/or gastrotomy, thoracoabdominal approach and end to end anastomoses of the proximal and distal atresia. [4,5,6]

Case Report

Here we present a case of a neonate who was delivered by a 29 years old primiparous mother at his 39+5 weeks of gestation by Caesarian section for non reassuring fetal heartbeat at a tertiary hospital with an Apgar score score of 8 and 9 in the 1st and 5th minutes of delivery. Two hours after delivery the neonate developed severe shortness of breath which was progressively

worsening. The baby was immediately referred to our hospital. On admission, the physical examination revealed decreased air entry on the right side of the chest. An urgent Chest x-ray done showed a right pneumothorax with anterior herniation of the right lung (Figure 1).

With the diagnosis of spontaneous tension pneumothorax, a chest tube was inserted with gush of air with subsequent decrease in the respiratory distress. Thereafter, the baby started to have profuse salivation and chocking episodes even though the baby had not been started on oral feeding because of the severe respiratory distress. Consequently, a diagnosis of an esophageal atresia (EA) with tracheoesophageal fistula (TEF) was made. A nasogastric tube was inserted. A repeat PA chest x-ray taken showed recoiling of the NG tube at around T6 vertebra (Figure 2). Pre-operatively, the baby was investigated for any other associated anomalies but none was found.

Operative procedure

The baby was taken to theatre where a right thoracotomy at the 4th intercostal space was done. At operation, the TEF was identified (Figure 3), was isolated and ligated. The proximal atresia was identified overlapping the fistula, was mobilized and opened by trying to anastomose the posterior wall to the distal segment. When the NG tube was passed distally, it got stuck in the distal esophagus at T 8-9 level. When an esophagotomy was done about 1cm proximal to the holdup, a membranous atresia of the esophagus was identified which was resected. The esophagotomy was repaired and the NG tube pushed distally into the stomach. The proximal esophageal atresia anterior wall repaired and the haemostasis secured. The previously inserted chest drain was replaced with a new one. The chest wall closed in layers (Figure 3).

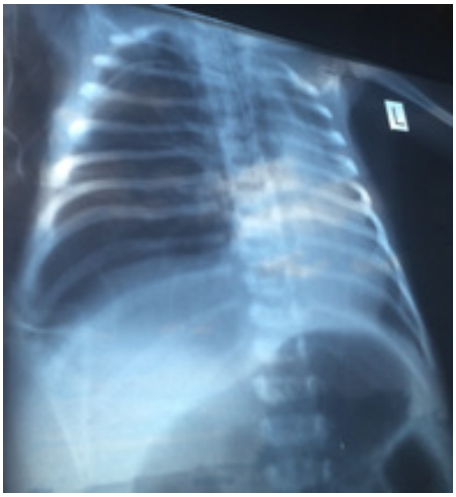
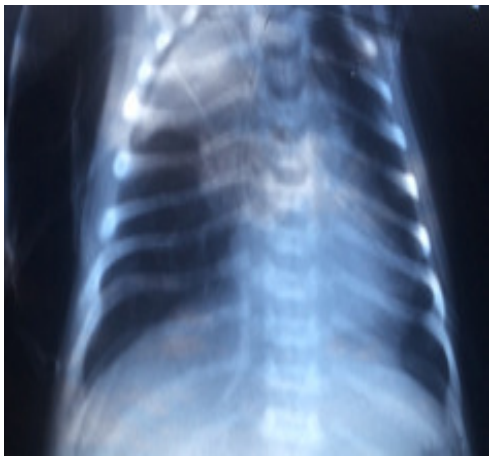


Figure 1. Tension Pneumothorax

Postoperatively the baby's respiratory distress progressively improving and at 4th postoperative day, the NG tube feeding was started. When the oral feeding was started on the 7th day, the neonate had mild leak seen through the chest tube and confirmed by contrast study (Figure 4) But with only conservative management it is by chest drain the leak stopped , the baby tolerated feeds & with total improvement he was discharged at his 1 month of age .(Fig 5).He had a good & stable outpatient follow up except contact dermatitis to his face which responded to topical management. And he started solid diet at his 6 months of age & tolerated well without dysphagia & become 10Kg at his 7 months of age.



2. Esophageal Atresia with Tracheoesophageal Fistula (TEF)

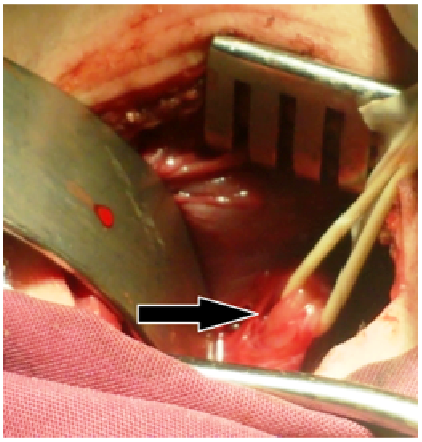


Figure 3. Intraoperative picture

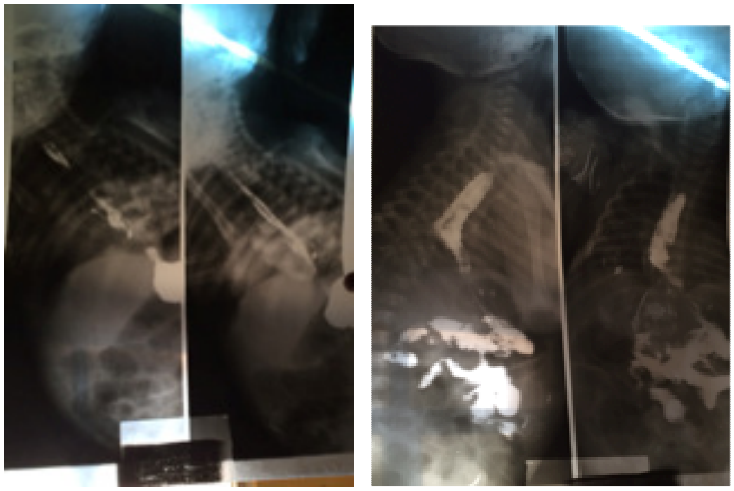


Figure 4. Contrast study Leak

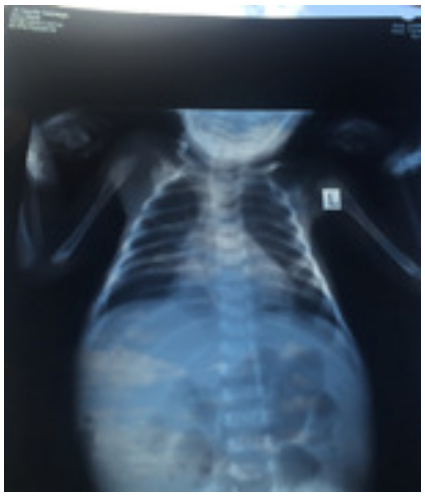


Figure 5. Post operative before discharge

Discussion

Spontaneous pneumothorax can occur in the newborn infant and if not recognized may have fatal consequences. It should be suspected in any infant in respiratory distress. Spontaneous pneumothorax is probably due to occlusion of some of the smaller air passages by aspirated meconium. The incidence of its occurrence is high in preterm infants with less mature lung than term infants especially for those who are admitted to NICU & put on mechanical ventilators than a term neonate that is 1-2% of the total cases [1,2]. The patient with a small pneumothorax and only moderate distress may recover completely with no treatment other than observation in an oxygen-enriched atmosphere. If considerable distress persists, continuous drainage of the pneumothorax should be provided by means of an intercostal drain and an underwater seal [1]. Our neonate had developed the respiratory distress 2 hours after the delivery while he was at the side bed of his mother for which the specific cause was not known that is why we said spontaneous. But since he was in distress and the pneumothorax was causing tension our mode of management was chest tube drain by 12 Fr tube to which treatment the baby improved.

The diagnosis of EA easily made by relatively rigid radio-opaque 6 Fr NG or bigger size i.e 10Fr OG tube insertion and taking plain chest x-ray. If there is an atresia there will be a resistance in pushing the tube and on the x-ray, the coiling of the tube will be seen. Additionally on the X-ray we will look for abdominal gas shadow to know the association with distal TEF which shows gas at the stomach site or in the abdomen which is not visible in pure atresia or with proximal TEF. In our case we saw the coiling go the NG tube at T6 vertebra which mean the proximal atresia is somehow down i.e the gab to the distal will be short and will be easy for the anastomoses [3].

But since it is rare suspicion of double atresia can not be made preoperatively with any means of investigation but intraoperatively failure of passage of the NG or OG tube distally will make you suspect the diagnosis and confirmed by opening the oesophagus on the second site of resistance [7]. In our case the resistance of NG tube advancement occurred at T8-9 where esophagotomy done and confirmed.

The distal atresia usually is membranous type & also there are some reports of distal membranous oesophageal atresia which are just above or below the diaphragm but they are single atresias and mostly without TEF. The management of the distal membranous atresia was easy i.e by esophagotomy or gastrostomy and identifying the membrane, resecting out and

repairing the oesophagus or the stomach[4,5,6]. Here we do it through opened oesophagus.

Conclusion

Even if spontaneous pneumothorax is not the commonest manifestation of EA with TEF , it can happen in any neonate. It should be suspected in any infant in respiratory distress. And although Gross and Vogt doesn't describe double oesophagealatresia ,it can be the variant with or without TEF.

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