Fistula formation between trachea and the innominate artery is a life-threatening complication of tracheostomy. The overall incidence of tracheo-innominate artery fistula (TIF) has been reported to be 0.3-0.7%. Several such cases occur in the first three weeks after tracheostomy and its hallmark is massive arterial bleeding either through or around the tracheostomy site. Mechanisms of fistula formation include mucosal necrosis due to pressure exerted by the cuff of the tracheostomy tube, high-cuff pressure, abnormally high innominate artery, low-placed tracheostomy (4 tracheal ring or below) and excessive head movement. Immediate surgical attention is the key to managing this fatal complication.

We present herein patient who developed TIF and died as a consequence of massive haemorrhage into the tracheobronchial tree and asphyxia. It is a rare case and to our knowledge no case of TIF or any case series concerning this rare complication has recently been reported from Pakistan.

Keywords: Tracheo-innominate fistula, Organophosphate poisoning, Tracheostomy complication.

Introduction
The use of tracheostomy for respiratory support of critically ill patient is a lifesaving form of therapy. One of the rare complications of tracheostomy is fistula formation between the trachea and the innominate artery. This case is being reported on account of its rarity.

Case Report
A 24-year-old married woman was admitted to the Intensive Care Unit (ICU) after the ingestion of half a bottle of a compound containing organophosphate 10 hours earlier with intent of self-harm. She was admitted with signs and symptoms of cholinergic crisis. The condition was managed by giving atropine which was continued as per the requirement. Injection pralidoxime 1gm, q8 hourly, and injection ceftriaxone 1gm, twice daily, was also given.

The next day, the patient became apnoeic and hypoxic, requiring emergency endotracheal intubation and mechanical ventilation. She was fed through a nasogastric tube and anti-cholinergic treatment was continued. On the fifth and seventh day of admission, the patient’s endotracheal tube was found to be blocked due to clot formation and the endotracheal tube (ETT) was changed accordingly. This was followed by subcutaneous emphysema, which resolved within 24 hours.

On the eighth day, tracheostomy was performed due to continued ventilation without any complications. Five days after tracheostomy, the patient was able to breathe on her own. Atropine was tapered and the antibiotic was also stopped as the patient became afebrile.

Ten days after tracheostomy, while still in the ICU, the patient started bleeding through the tracheostomy tube. It was sudden, projectile, bright red, high-pressure blood emerging from the tracheostomy tube opening. The anaesthetist present in the ICU immediately removed it and ETT was passed with its balloon inflated soon after the insertion. Around one litre of blood was suctioned from the respiratory tract in addition to approximately two litres that she had bled earlier. Colloids, intravenous fluids and unmatched blood were given in an effort to resuscitate the patient, but she died within 30 minutes.

Discussion
Bleeding is a common complication of tracheostomy and its incidence varies from 1-3%. The common causes of early post-tracheostomy haemorrhage include inadequate surgical homeostasis and coagulopathy. However, massive delayed haemorrhages occur rarely after tracheostomy and can be serious. The incidence varies from 0.6-0.7%. The common causes of massive delayed haemorrhage includes granulation tissue formation, tumour invasion, pulmonary artery rupture.
and TIF. The innominate artery is involved in 80% of the cases due to its close anatomic relation to the anterior surface of trachea (Figure-1). Fistula formation with the common aortic arch, common carotid artery, superior and inferior thyroid artery and innominate veins have also been reported.

TIF is a rare but devastating complication of tracheostomy, which usually presents with acute tracheal bleeding. It has been reported to occur in less than 1% of the patients and only those patients who receive urgent surgical intervention are able to survive; otherwise the mortality rate approaches 100%.

The formation of TIF is multifactorial. It involves the destruction of the anterior wall of the trachea and the posterior wall of the innominate artery. The precipitating factors, which have been implicated for TIF formation, are mucosal necrosis from high-cuff pressure, mucosal trauma from malpositioned cannula tip, low tracheal incision and hyperextension of the neck. Factors other than anatomical considerations also play an important role in tracheal erosion. The presence of pathological organism can also be responsible for the degeneration of tracheal structures. Long duration of tracheostomy and inadequate attention to septic techniques during aspiration and dressing provides a favourable ground for organisms to cause destruction. Devitalised tracheal tissue already present at the time of conversion to a tracheostomy can further contribute to massive destruction. This is also applicable to this case as during the tracheostomy, the anatomy of tracheal rings was found to be distorted. From the above factors, the possible reasons for TIF formation applicable to our case include: damage to tracheal rings probably during one of the ETT intubation (as subcutaneous emphysema was noted after one such attempt); trauma to the trachea due to hyperinflation; exhaustive neck movements which may have occurred as the patient was restless; and vigorous tracheal suctions and pulmonary toilet.

Three cardinal signs can help in the diagnosis of TIF: stomal bleed, haemoptysis or any pulsations felt in the tracheostomy tube. In this patient, the ETT was blocked (due to clot formation) a few times before the final event took place. However, pulsation was not observed. Pulsation of tracheostomy tube has only been reported in 5% of the patients who were later found to have had TIF.

A differential diagnosis for the attending clinician is based on the time lag between the tracheostomy and subsequent haemorrhage. Haemorrhage occurring 3 days to 6 weeks after tracheostomy should always be taken as a red flag for TIF. In this patient, massive haemorrhage occurred 10 days after tracheostomy, which was suggestive of TIF. Other causes of TIF formation include thoracic aneurysm rupture and vascular fistula. Moreover, 70% of all delayed haemorrhages occur during the first three weeks; this is also consistent with our case. Approximately 50% of the reported cases that have had minor, bright red, preliminary (sentinel) bleed later develop massive delayed haemorrhage. Sentinel bleeding is reported to be self-limiting but recurrent in nature and is aggravated by coughing or aspiration. When TIF is suspected, the patient must immediately be taken to the operation room where bronchoscopy and slow-cuff deflation must be performed simultaneously, followed by gradual removal of the tracheostomy tube. The use of bronchoscopy is recommended to confirm the extent and source of bleeding. In our case, bronchoscopy was not available at the time of the event.

The management depends on the speed and volume of haemorrhage (Figure-2). If brisk haemorrhage has occurred, tracheostomy tube cuff hyperinflation may help to control the bleeding temporarily. However, if
Figure-2:

massive haemorrhage occurs with high suspicion of TIF, as in our patient, orotracheal tube can be inserted distal to the bleeding site and the tracheostomy tube should be removed simultaneously. This procedure can help to secure the airways and will provide an airway for diagnostic bronchoscopy. It can also prevent asphyxia. If the above measures fail, digital compressions of the innominate artery through the tracheostomy tract should be performed. Digital compression consists of inserting fingers into pretracheal space to tamponade the innominate artery against the manubrium. The digital compression should be maintained during the transport to the operation room until surgical control is achieved. This procedure has shown to terminate bleeding in 90% of the patients.

Prevention is the best treatment. Thus, the key to prevention of TIF includes avoidance of unnecessary tracheostomies, proper tracheostomy care, and preventing tracheal damage by avoiding excessive head movements and prolonged over-inflation of the cuff.

**Conclusion**

Management of bleeding from tracheostomy is a difficult task requiring a multi-disciplinary team experienced in dealing with such complications. They should be present in all ICUs. We recommend that all doctors and nursing staff involved in the ICU care should have awareness of, and training in, tackling this emergency situation.

**References**