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Penetrating chest injury in a case of situs inversus totalis

Nasirudeen Oladipupo Ajayi[1,2], Lelika Lazarus[1], Kapil Sewsaran Satyapal[1,*]

[1] Department of Clinical Anatomy, School of Laboratory Medicine and Medical Sciences, College of Health Sciences, University of KwaZulu-Natal, Westville Campus, Durban, South Africa
[2] Benedictine Hospital, Nongoma, KwaZulu-Natal, South Africa

Abstract

Situs inversus totalis is a congenital condition characterized by the transposition of the thoracic and abdominal organs to the opposite side of the body. *Situs inversus totalis* is typically asymptomatic, however, these individuals are susceptible to certain defects and malformations such as vascular anomalies and mal-positioned viscera, which can result in a shortened lifespan. There are reports on penetrating abdominal injury in patients with *situs inversus*. In addition, the presence of pathology of thoracic and abdominal organs in *situs inversus* patients have been reported. However, from the literature reviewed, there is a paucity of reports on penetrating chest injuries in patients with *situs inversus*. Hence, we present a case of a patient who presented with a stab chest with *situs inversus totalis* and a brief discussion on the embryology of the condition.

Key words

Chest injury, situs inversus totalis, embryology.

Introduction

Thoracic and abdominal organs such as the lungs, atria, liver, spleen and stomach are commonly asymmetric. The position of these organs relative to the midline is described by the term situs solitus (Gindes et al., 2007). Historically, a case of the reversal of the liver and the spleen was reported by Fabricius in 1606 and the transposition of the viscera was first recognized by a clinician in 1824 (Cleveland, 1926). Situs inversus totalis (SIT) is a congenital condition characterized by the transposition of the thoracic and abdominal organs to the opposite side of the body (Nakano et al., 2017). This anomaly can be inherited through an autosomal recessive trait, however, the precise genetic cause is indeterminate (Nursal et al., 2001, Suh 2017). SIT can exhibit dextrocardia which is characterized by the reversal of the chambers of the heart (Suh 2017). In 90-95% of cases presenting dextrocardia with SIT, the heart is structurally normal (Marta et al., 2003). This is in contrast to dextrocardia with situs solitus referred to as dextroversion, which reports a higher frequency in structural defects (Marta et al., 2003). SIT is also characterized by abdominal organs such as the stomach and spleen located on the right side, as well as the liver and gallbladder located on the left side of the midline (Suh, 2017).

* Corresponding author. E-mail: satyapalk@ukzn.ac.za

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Situs inversus totalis can vary in incidence from 1 in 5000 to 20 000 individuals, and this is possibly as a result of different diagnostic methods (Oms and Badia, 2003; Uludag et al., 2017). People presenting with SIT are typically asymptomatic, however, these individuals are susceptible to certain defects and malformations such as vascular anomalies and mal-positioned viscera, which can result in a shortened lifespan (Sugawara et al., 2001; Giuliani et al., 2017). The anatomical variations presented in this condition can cause significant technical difficulties during surgical treatment, regardless of there being no obvious abnormalities in the functioning of the transposed organs (Uludag et al., 2017). The difficulty in diagnosis and surgical management of patients with SIT is a result of the contralateral disposition of the viscera (Oms and Badia, 2003). Procedures such as ultrasonography or auscultation of the heart during physical examinations may contribute to the accurate diagnosis of SIT (Nursal et al., 2001).

The occurrence of complete situs inversus has no gender or racial disparity. It is most often an isolated and accidental event occurring in an individual and rarely runs in families (Supriyah et al., 2013). There are reports on penetrating abdominal injury in patients with situs inversus. In addition, the presence of pathology of thoracic and abdominal organs in situs inversus patients have been reported. However, from the literature reviewed, there is paucity of reports on penetrating chest injury in a patient with situs inversus. Hence, we present a case of stab chest in a patient with SIT and a brief discussion on the embryology of situs inversus.

Case report

A 32 year old male presented to the emergency room of a rural hospital in South Africa approximately 10 hours after sustaining a stab wound to the chest. On examination he was found to have 3 cm deep laceration inferior to the middle third of the left clavicle. On clinical examination, he was not in respiratory distress; the trachea was centrally located, there was good air entry bilaterally on auscultation of the lungs. He was hemodynamically stable. Blood pressure and laboratory blood results were within normal limits. He had an erect chest x-ray which showed clear lung fields without hemo-pneumothorax but an incidental finding of dextrocardia and the location of the gastric bubble on the left of the abdomen was observed (Figure 1). The laceration was irrigated, cleaned and sutured. He was subsequently admitted for observation due to the depth of the laceration. An ultrasound of the abdomen revealed the transposition of the abdominal organs with a right-sided spleen. The liver and gall bladder were left-sided with the liver having a normal echotexture; the gall bladder presented with no calculi. A repeat chest x-ray 24 hours after admission to the hospital revealed a collapsed left lung with a hemo-pneumothorax (Figure 2). The patient was administered with a left thoracostomy tube for the management of the left hemo-pneumothorax and about 200 ml of blood were drained. A post-thoracostomy chest x-ray revealed the expansion of the left lung with visible left costophrenic angle (Figure 3). The thoracostomy tube was removed and the patient was counseled and discharged without any complication.

Table 1. Incidence of SIT.

Author (year)	Incidence of SIT
Applegate et al. (1999)	0.01% of general population
Nursal et al. (2001)	1:5000 20 000 hospital admissions
Sugawara et al. (2001)	0.025% to 0.005% of general population
Marta et al. (2003)	1:10 000 births
Oms and Badia (2003)	1: 5000 to 20 000 people
McKay and Blake (2005)	1:5000 – 20 000 people
Kouwenhoven et al. (2007)	1:10 000 of general population
Ramling and Dakshayani (2014)	1:10 000 live births
Giuliani et al. (2017)	1:5000 to 1:10 000 adults
Juma et al. (2017)	1-2 per 10 000 individuals
Nakano et al. (2017)	0.005% - 0.01% live births
Segel (2017)	6000 – 8000 live births
Suh (2017)	1:10 000 to 50 000 persons
Uludag et al. (2017)	1:5000 to 20 000 people

Discussion

Anatomic asymmetry is established during embryogenesis, and the left-right axis is established during early embryological development (Marta et al., 2003). Positioning of the organs and determining their asymmetries is orchestrated by a cascade of signal molecules and genes such as LEFTY, NODAL, IV (inversus viscerum), HAND, ZIC3, SHH, ACVR2B, and PITX2 genes (Marta et al., 2003; Sadler 2015). The gene PITX2 is responsible for determining left-sidedness and if expressed ectopically can lead to laterality defects (Sadler, 2015). The genes that influence the development of the right side are not as clearly described, however, the transcription factor SNAIL is thought to control the genes responsible for establishing right-sidedness (Sadler, 2015).

Situs inversus totalis is a laterality defect that occurs during embryological development (Uludag et al., 2017). This can be a result of random developmetal events, genetic or environmental factors (Catana and Apostu, 2016; Segel, 2017). Suh (2017) suggested that the development of SIT is due to immobility of the nodal cilia which inhibits the flow of extra-embryonic fluid during embryogenesis. Trulioff et al. (2017) suggested that the assumption of two types of cilia can provide a possible explanation for the role of cilia in SIT. This hypothesis assumes that there are movable primary cilia which generate and rotate extra-embryonic fluid flow, as well as immobile cilia which serve as mechanoreceptors of the flow and can influence the expression of the genes responsible for left-sided development (Trulioff et al., 2017).

Many people with SIT are oblivious of their uncommon anatomy until they seek medical attention for an unrelated condition (Supriyah et al., 2013). In the majority of cases (90-95 %), the presence of situs inversus with dextrocardia and normal anatomic relationship of the great vessels is associated with normal cardiac structure and

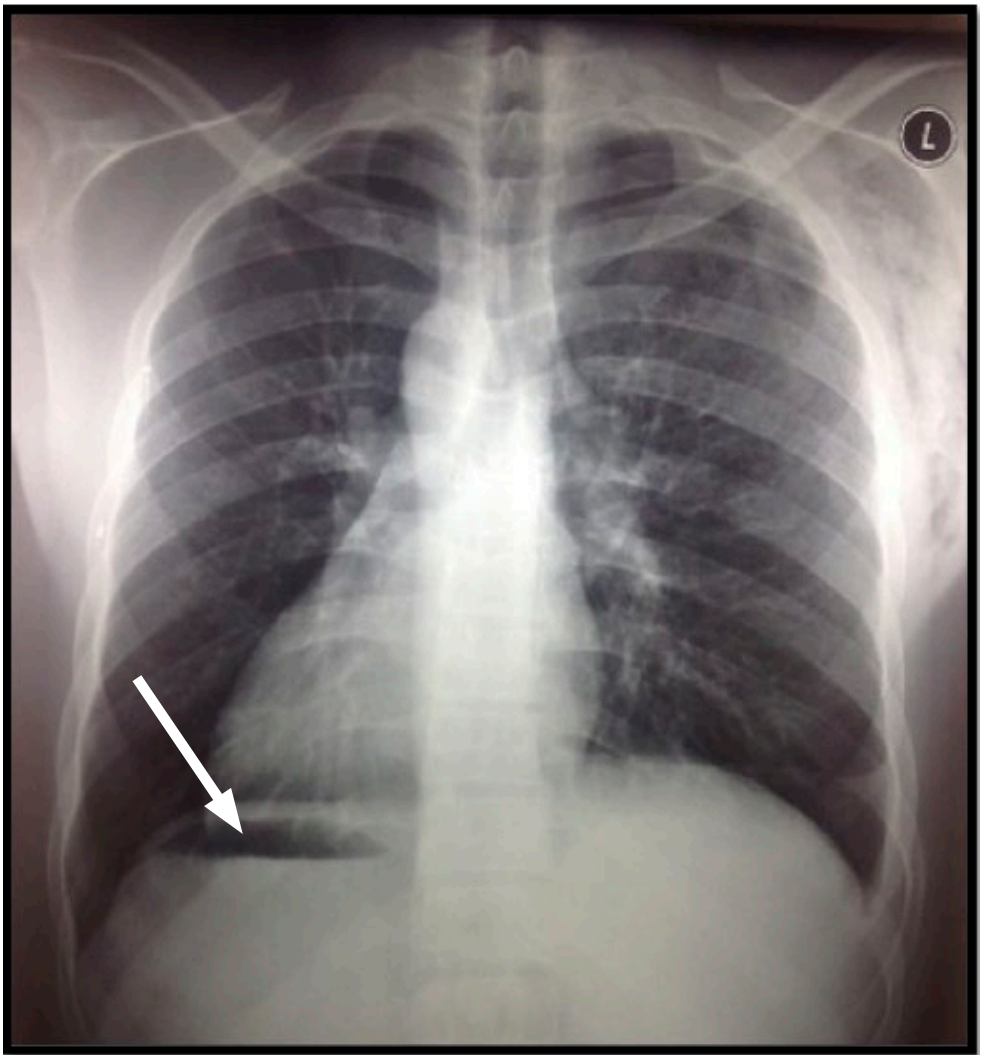

Figure 1. Chest X-ray (posteroanterior view) showing heart in the right hemithorax and the gastric bubble on the right (white arrow).

function. However, congenital cardiac disease such as tetralogy of Fallot or pulmonary artery atresia and interatrial or interventricular septal defects are not uncommon (5-10% of cases) (Marta et al., 2003). Therefore, most patients with situs inversus totalis live a normal life.

The presence of the combination of situs inversus, dextrocardia, bilateral cystic bronchiectasis and chronic sinusitis confirms the diagnosis of Kartagener's syndrome in these patients. (Mohan et al., 2007). Situs inversus may be associated with other congenital anomalies such as duodenal atresia, asplenism, multiple spleens, ectopic

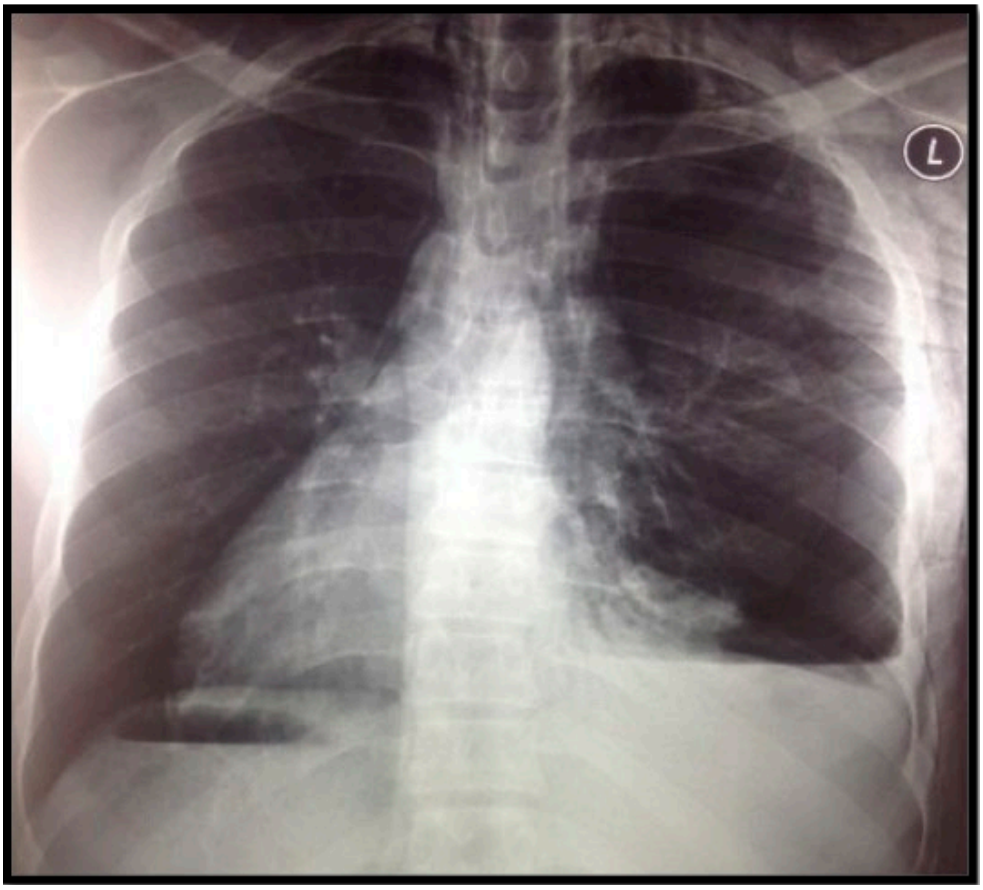

Figure 2. Chest X-ray (posteroanterior view) showing left hemo-pneumothorax.

kidney, horseshoe kidney and various pulmonary and vascular abnormalities (Kumar et al., 2014). On abdominal ultrasound, the patient in this case report had the liver and spleen transposed and there were no gross anomaly of the abdominal organs.

Penetrating trauma in the proximity of major vessels may result in vascular injury and the prompt detection of such injuries will facilitate early treatment and minimize complications (Williams et al., 2007). The clinical presentation in patients with situs inversus differs from what is found in most patients because the organs are a mirror image of the usual anatomy. The presence of situs inversus may prevent the occurrence of severe vascular injury or may be deleterious in some cases. In the present case report, the location of the stab to the left side of the chest could have affected some vital structures such as the arc of the aorta or the heart. However, the presence of dextrocardia and 'inversion' of the thoracic organs may have protected the patient from serious vascular injury had the penetration been very deep or lower in the thoracic cavity. Therefore, in cases where the clinical presentation does not correlate with

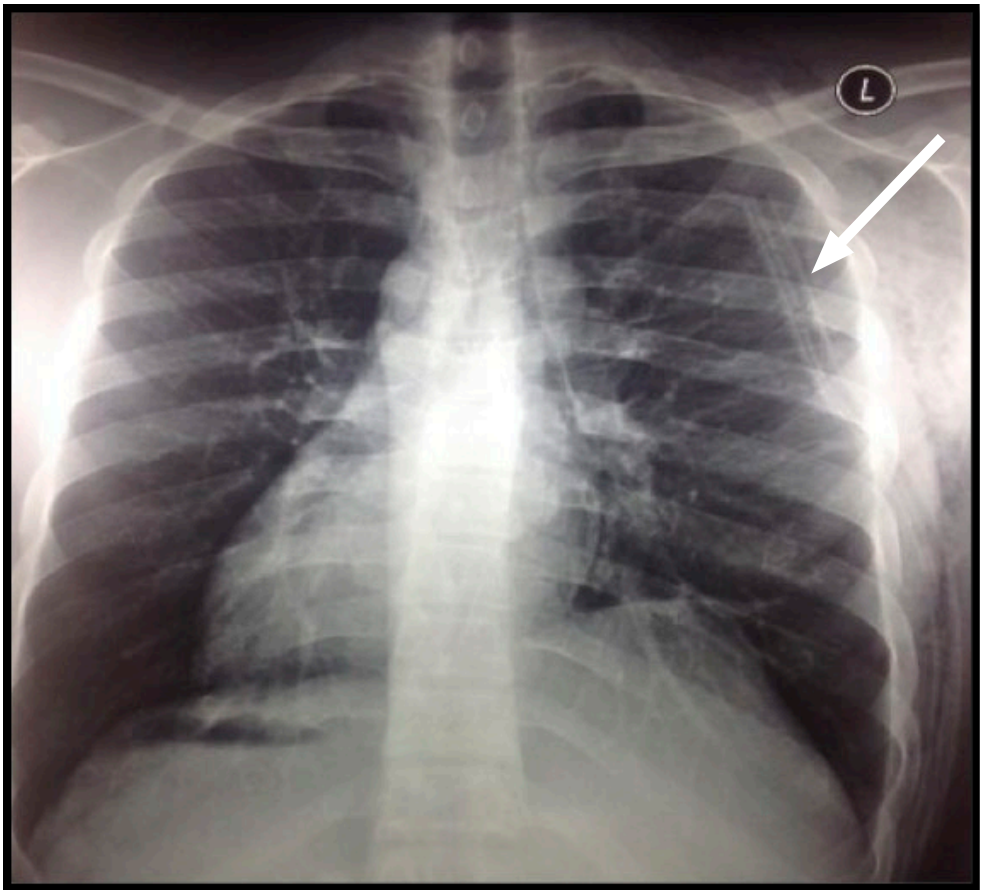

Figure 3. Chest X-ray (posteroanterior view) showing re-expansion of the left lung and resolution of the hemo-pneumothorax with the chest tube (white arrow) in-situ.

the location of the injury or suspected pathology, an unusual anatomy of the thoracic or abdominal organs should be suspected.

The patient in this report was successfully managed with a thoracostomy tube drainage for his hemo-pneumothorax which does not differ to the management for a situs solitus patient. Surgical procedures can be performed successfully in situs inversus, however, such operations are technically challenging (Ahmad et al., 2015). The identification of situs inversus is important for preventing surgical complications that could result from the inability to recognize the reversed anatomy (Casanova et al., 2008). Due its rarity, situs inversus may complicate organ transplantation procedures as donor organs will most likely come from situs solitus donors (Supriyah et al., 2013).

Conclusion

Penetrating thoracic injury in a situs inversus patient could be safely managed as in patients with situs solitus. The 'inversion' of the thoracic organs maybe of an advantage as it may preclude severe vascular or organ injury.

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