SPONTANEOUS PNEUMOMEDIASTINUM: A RARE AND UNUSUAL COMPLICATION OF DIABETIC KETOACIDOSIS

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Abstract

Spontaneous pneumomediastinum is defined as the presence of free air in the mediastinum without any apparent concomitant factors or disease. It is most often affecting young males which is usually benign and self-limiting. The pathophysiology of this disease is probably based on a pressure gradient between the alveolus and the lung interstitium. The most important examination to make a diagnosis of spontaneous pneumomediastinum is radiography. Generally, no special interventions are indicated for the treatment. We report a case of 26-year-old man with a benign spontaneous pneumomediastinum complicating an inaugural diabetic ketoacidosis.

Introduction:

Spontaneous pneumomediastinum (SPM) is defined as a pneumomediastinum that is not related to trauma, surgery, or other medical procedures. This entity is usually precipitated by high intrathoracic pressure conditions, such as vomiting, labor, or sneezing. [1]

SPM as a complication of diabetic ketoacidosis (DKA) is very rare and is likely related to vomiting. The following case illustrates a diagnostic and management challenge in a young male patient presenting with inaugural DKA, complicated by SPM.

Case Report:

A 26-year-old man with no past medical history presented to the emergency department with three days of vomiting, abdominal pain, and progressive difficulty of breathing.

On clinical examination, the patient was dehydrated, polypneic at a rate of 50 breaths/min, tachycardic at 120 bpm and his blood pressure was 140/99 mm Hg. The temperature was normal at 37°C and oxygen saturation 93 % in room air. The lung, heart, and abdomen exam were unremarkable and there was no neck crepitus noted.

The laboratory evaluation showed leukocytosis of 21 x1000/mm³, elevated creatinine of 2 mg/dL, hyperglycemia of 780 mg/dL, hyponatremia of 130mEq/L, hyperkalemia of 5.2 mEq/L, and bicarbonate of 5 mEq/L. The anion gap was 27 and a venous blood gas showed a pH of 6.7. Urinalysis was positive for 3+ ketones and 1+ protein.

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A chest X-ray on admission showed free air in the mediastinum outlining the left and right heart border without pneumothorax (Fig 1).

Chest computed tomography (CT) reaffirmed the presence of extensive pneumomediastinum involving the base of the neck, around the great vessels, around the esophagus, and around the pericardium. There was no pneumothorax or any finding on lung parenchyma (Fig 2). Oesophageal contrast studies showed no perforation of the oesophagus.

A diagnosis of diabetic ketoacidosis (DKA) with pneumomediastinum was made.

The patient was admitted to the intensive care unit for monitoring. His ketoacidosis resolved within 24 hours following initiation of intravenous normal saline, bicarbonate, and insulin drip as per DKA protocol. For pneumomediastinum, he was treated conservatively with analgesia and respiratory support.

The patient was discharged home after 5 days without any complications with a plan for early review at the diabetes day centre.

Discussion:-

Pneumomediastinum (PM) is a rare condition characterised by the presence of free air in the mediastinum. It is due to various conditions leading to alveolar overdistension and barotrauma. These can result in alveolar rupture and as a result, air leaks into the pulmonary interstitium, dissecting through the bronchovascular bundles into the mediastinum.[1]

Air canalsotrack up into the pleural space, subcutaneous soft tissues of the neck and pericardium leading to pneumothorax, soft tissue emphysema and pneumopericardium, respectively.[2]

Spontaneous pneumomediastinum (SPM) is defined as that occurring without surgical or medical procedures, chest trauma or mechanical ventilation. In 1937, Hamman described SPM for the first time.[3] Since that date, many cases were reported in conditions leading to high intrathoracic pressure swings such as coughing, straining, vomiting, strenuous crying especially in people with asthma or chronic obstructive pulmonary disease. But only a few case reports have described the association with DKA.[4]

The exact pathophysiology is still unknown; however, it is believed that Kussmaul respiration leads to a significant 20–30 mm Hg rise in intra-alveolar pressure and this may result in alveolar rupture. Furthermore, vomiting can predispose to alveolar rupture through increasing intrathoracic pressure.[5]

SPM can be asymptomatic; however, chest pain, shortness of breath, and subcutaneous emphysema have been reported. Hamman’s sign is an infrequent clinical sign in the presence of mediastinal emphysema and is heard as a crunching sound over the precordium, synchronous with the heart beat.[6,7] Our patient did not present with emphysema on physical exam.

The diagnostic imaging for SPM is a two-view chest X-ray, which has a sensitivity of 50%-90%. When suspicion is high and the chest X-ray is negative, a CT chest without contrast could be the next step in evaluation.[6] The features of the X-ray are generated by leaked air itself and an enhanced margin of mediastinal structures by the air. On the posterior-anterior view of the chest X-ray, the commonest three findings are the following: air streaks in the superior mediastinum (sometimes they reach to the neck), the prominent silhouette of the heart (especially on the left) and subcutaneous emphysema of the shoulder and neck. Infrequent but characteristic features include the double-bronchial wall sign (the visualized tracheal or main bronchial wall putted between inner air and leaked air) and the continuous diaphragm sign (the diaphragm of both sides appearing connected by leaked air between the inferior surface of heart and diaphragm). Especially in paediatric cases, the thymic spin maker sailsign is produced by airlifting the thymus off the mediastinal structures.[8] On chest CT, the leaked air mainly distributes from the anterior mediastinum to the neck, and the amount of air is often more than estimated on a chest X-ray. The air may extend to the pericardium, retroperitoneum, peritoneum or spine.

Because chest CT is essential to detect small air leakages, it should be performed to make a correct diagnosis in strongly suspicious cases of SPM, even if the chest X-ray is in the normal range.[9] Furthermore, chest CT may reveal other findings such as pre-existing pulmonary disease.
Boerhaave’s syndrome is rarely associated with SPM; however, it is important to keep it in consideration due to its associated high mortality rate of 70% [5]. This can be suspected in a patient presenting with severe emesis with or without blood leukocytosis, hypotension, and a prior history of gastroesophageal reflux disease. CT chest without contrast or esophageal swallow imaging can be useful diagnostic tools [6]. Recurrence of SPM is extremely rare [10].

As evidenced in our case, SPM is generally a benign self-limited condition. Management is generally conservative, including rest, oxygen, and analgesia [11]. There is no established role for antibiotics unless there is a concomitant infectious process, neither is there a specific follow-up indication for patients who had an uncomplicated SPM. Fortunately, the evolution of this entity is benign with an excellent prognosis and no recurrence in most cases [12].

Figure:-

**Figure 1:** A chest X-ray showing free air in the mediastinum outlining the left and right heart border without pneumothorax (white arrows).

**Figure 2 (a,b):** Chest computed tomography without contrast showing the presence of pneumomediastinum involving the base of the neck (black arrow), around the great vessels, around the esophagus, and around the pericardium (white arrows).
Conclusion:
SPM is a rare and unusual complication of DKA which results from the increased intra-alveolar pressure and alveolar rupture due to vomiting and ketotic breathing associated with DKA. It is usually asymptomatic and resolves with conservative management in most cases.

Conflict of interest statement:
Authors declare that there is no conflict of interest.

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References:
1. Abolink I, Lossos IS, Breuer R. Spontaneous pneumomediastinum. A report of 25 cases. Chest (1991);100:93–5.
2. Panacek EA, Singer AJ, Sherman BW, et al. Spontaneous pneumomediastinum: clinical and natural history. Ann Emerg Med (1992);21:1222–7
3. Guzman rojas P, Agostinho J, Hanna R, et al. Spontaneous Pneumomediastinum as a Consequence of Severe Vomiting in Diabetic Ketoadicosis. Cureus (2018) 10(5): e2562. DOI10.7759/cureus.2562
4. Pauw RG, van der Werf TS, van Dullemen HM, et al. Mediastinal emphysema complicating diabetic ketoacidosis: plea for conservative diagnostic approach. Neth J Med (2007);65:368–71.
5. Pooyan P, Puruckherr M, Summers JA, et al. Pneumomediastinumpneumopericardium, and epidural pneumatosis in DKA. J Diabetes Complications (2004);18:242–7.
6. Lane AS, Champion B, Orde S, Dravec D: Diabetic ketoacidosis due to fulminant type I diabetes: a rare subtype of type I diabetes leading to unusual sequelae. J Intensive Care Soc. (2015), 16:64-70. 10.1177/1751143714551249
7. Girard DE, Carlson V, Natelson EA, Fred HL: Pneumomediastinum in diabetic ketoacidosis: comments on mechanism, incidence, and management. Chest. (1971), 60:455-459.10.1378/chest.60.5.455
8. Bejvan, S.M. and Godwin, J.D. Pneumomediastinum: old signs and new signs. AJR (1996)166: 1041_1048.
9. Iyer, V.N., Joshi, A.Y. and Ryu, J.H. Spontaneous pneumomediastinum: analysis of 62 consecutive adult patients. Mayo Clin Proc (2009) 84:417_421.
10. Natale C, D’Journo XB, Duconseil P, et al. Recurrent spontaneous pneumomediastinum in an adult. Eur J Cardiothorac Surg (2012);41:1199–201.
11. Steenkamp D, Patel V, Minkin R: A case of pneumomediastinum: a rare complication of diabetic ketoacidosis. Clin Diabetes. (2011), 29:76-77. 10.2337/diabetes.29.2.76
12. Macia I, Moya J, Ramos R, et al.: Spontaneous pneumomediastinum: 41 cases. Eur J Cardiothorac Surg. (2007), 31:1110-1114. 10.1016/j.ejcts.2007.03.008.