

Spontaneous Pneumorachis – A Case-Based Review

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Abstract: Pneumorachis is characterized by the presence of free air in the spinal canal. It is referred by different names in literature such as epidural emphysema, intraspinal air, intraspinal pneumoc(o)ele, spinal epidural and subarachnoid pneumatosis, spinal and epidural emphysema, aerorachia, pneumosaccus, air myelogram, etc. Pneumorachis can be broadly classified as traumatic, iatrogenic, or spontaneous. In this case-based review, we present a case of spontaneous pneumorachis secondary to asthma exacerbation. This is followed by a systematic review of all cases of spontaneous pneumorachis identified in PubMed. The aim of this review is to understand the pathophysiology, common causes and the management of spontaneous pneumorachis.

Keywords: pneumorachis, spontaneous pneumorachis, marijuana smoking, asthma exacerbation

Introduction

Pneumorachis is a clinical condition characterized by the presence of free air in the spinal canal.¹ The presence of air in the cranial cavity, termed pneumocephalus, is a well-known clinical entity associated with skull fractures; on the other hand, pneumorachis is relatively rare. As early as 1918, air was utilized as a contrast medium for neuroimaging in a technique known as air myelogram. With the advent of less toxic contrast agents, the practice slowly died out. The first case of non-iatrogenic free air in the spinal canal (cervical) was reported in 1977 in a 20-year-old male who sustained multiple skull fractures during a motor vehicle accident. The clinical entity was initially described as “traumatic air myelogram.”³⁰ The current term pneumorachis was coined by Newbold et al in 1987.³¹ The condition is rare, and an accurate incidence rate is not available. As per our review, 48 case reports of spontaneous pneumorachis in adults have so far been published in the literature.

Based on the location of the air within the spinal cord, pneumorachis can be classified as intradural or extradural.⁹⁰ Extradural pneumorachis is more common and is usually benign. Intradural pneumorachis, on the other hand, is indicative of major trauma. While neurological deficits are rarely associated with pneumorachis, it is more commonly associated with intradural pneumorachis even though there are a few case reports of extradural pneumorachis causing nerve root compression or cauda equina syndrome requiring decompression.^{90,91}

Pneumorachis occurs most commonly due to either trauma or as a sequela of procedures involving the spinal canal.² Pneumorachis is termed spontaneous when it occurs in the absence of trauma or iatrogenic insult.³ Spontaneous pneumorachis is extremely rare, and only a few cases have been reported in the literature.⁴ Many of them have been reported in conjunction with asthma exacerbation, but there are

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no reports of marijuana-induced asthma exacerbation leading to spontaneous pneumothorax. The use of marijuana both for recreational and medicinal purposes is on the rise and the majority of current users smoke marijuana even when using it for medicinal purposes.⁹² Smoking marijuana is often associated with coughing paroxysms and is a potent trigger for asthma exacerbations making its use ideal for causing pneumothorax. With the increasing popularity of smoking marijuana and the widespread use of computerised tomography as a routine imaging modality, we believe clinicians are going to encounter spontaneous pneumothorax much more frequently in the future. In this article, we present a case of spontaneous pneumothorax and do a systematic review of spontaneous pneumothorax. Our goal is to consolidate the current understanding of this clinical entity so as to aid physicians to make the best informed clinical decisions.

Case Report

A 21-year-old male with a history of mild intermittent asthma presented with productive cough, sore throat, and neck pain for one day. The patient was smoking marijuana with his friends when he developed a sore throat, a severe bout of coughing with yellowish expectoration, post-tussive emesis, shortness of breath, and wheezing. He noticed a sharp, persistent pain starting in his neck and spreading to his anterior chest following his coughing paroxysm. He felt a sensation of swelling on his neck and could feel a crunching sensation when he touched it. He was not taking any home medications. He endorsed smoking a joint of marijuana daily and drinking 200 mL of hard liquor on weekends over the last 3–4 years.

On presentation, his blood pressure was 147/74 mm Hg, heart rate was 103 beats per minute, respiratory rate was 18 per minute, temperature was 98.7 °F, and he was saturating 94% on room air. His physical examination revealed extensive subcutaneous crepitus extending from his neck to the anterior chest wall. Auscultation of the lungs revealed diffuse expiratory wheezes with scattered rhonchi. Neurological examination did not show any motor, sensory or cranial nerve deficits.

Lab investigations are shown in **Box 1**. EKG revealed normal sinus rhythm. A chest X-ray showed pneumomediastinum along the left cardiac border and over the cardiome-diastinal silhouette anteriorly with extensive subcutaneous emphysema in the neck's soft tissues (**Figures 1 and 2**). A chest CT showed pneumopericardium and pneumomediastinum with air dissecting throughout the mediastinum, into the neck, down into the spinal canal, posterior paraspinal

Box 1 List of Laboratory Investigations

Laboratory Investigations		
Name	Result	Reference Range
White blood count	13,600 cells/mm ³	4000–11,000 cells/mm ³
Hemoglobin	17.3 g/dL	12.5–17 g/dL
Platelet count	163,000 cells/mm ³	150–450 × 1000 cells/mm ³
Sodium	140 mEq/L	134–144 mEq/L
Potassium	3.2 mEq/L	3.6–5.6 mEq/L
Chloride	102 mEq/L	96–109 mEq/L
Bicarbonate	24 mEq/L	20–32 mEq/L
Blood urea nitrogen	15 mg/dL	5–26 mg/dL
Creatinine	1.29 mg/dL	0.5–1.5 mg/dL
Glucose	118 mg/dL	65–99 mg/dL
Lactate	4.4 mmol/L	0.5–2 mmol/L
Phosphorus	3.1 mg/dL	2.5–4.5 mg/dL
C-reactive protein	68.28 mg/L	8–10 mg/L

musculature as well as adjacent chest wall musculature (**Figures 3 and 4**). Air in the spinal canal extended from C1 down to the T9 level. There was anterior midline pneumothorax extending from side to side, outlining the anterior and medial margins of the lungs. A transthoracic echo (TTE) showed a small amount of pneumopericardium.

There was no clinical or radiological evidence of tension pneumothorax. Since the patient was not in any respiratory distress, he was initiated on the treatment of asthma exacerbation with intravenous methylprednisolone 80 mg every 8

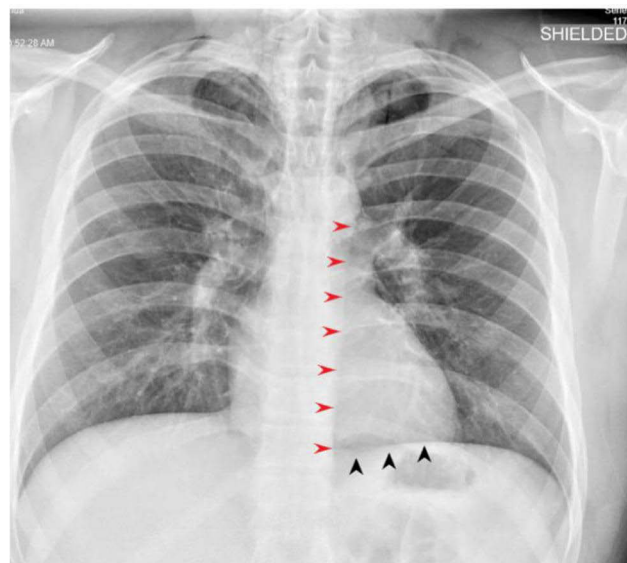


Figure 1 Chest X-ray PA view of the patient on the day of admission shows a "continuous diaphragm sign" characterised by a mediastinal gas outlining the superior surface of the diaphragm and separating it from the heart (black arrowheads) and a "Naclerio's V sign" in which mediastinal gas outlines the lateral margin of the descending aorta and extends laterally over the left hemidiaphragm (red arrowheads).

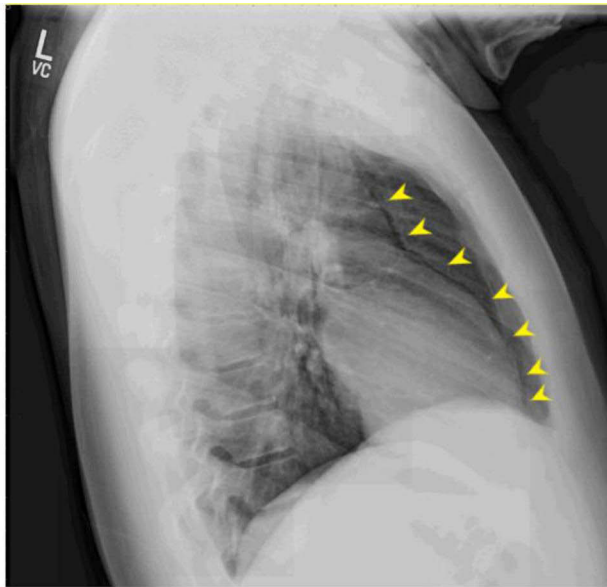


Figure 2 Chest X-ray (lateral view) demonstrating lucency (Yellow arrows) overlying the heart signifying pneumopericardium.

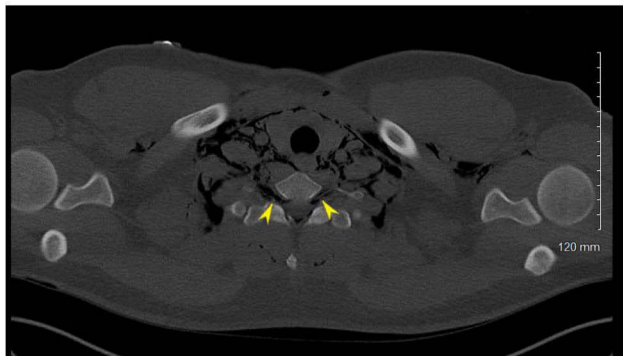


Figure 3 CT imaging demonstrating dissection of fascial planes in neck and invasion of trapped air into the spinal canal (yellow arrows) via intervertebral foramen.



Figure 4 Pneumorachis demonstrated in axial (black arrow) (A) and sagittal sections (B) of thoracic CT imaging (yellow arrow).

hours, budesonide nebulizers twice daily, albuterol nebulizers as needed, and oxygen through a nasal cannula. Daily chest X-rays and continuous pulse oximetry were obtained to monitor the pneumorachis. Chest X-ray on day 2 of hospitalization showed progression of subcutaneous emphysema.

On day 3 of hospitalization, the patient reported significant improvement in his symptoms. On physical examination, his wheezing and subcutaneous crepitus had resolved. Chest X-ray also showed resolution of subcutaneous emphysema and mild improvement in pneumomediastinum. His oxygen supplementation was weaned off. He was prescribed an albuterol inhaler as needed and discharged home. He was counseled on smoking cessation and educated on avoiding activities that increase intrathoracic pressure.

Systematic Review

Search Strategy

We utilized PubMed's search index for our systematic review. We used the search terms "pneumorachis" OR "spontaneous pneumorachis" under all fields to search for case reports and articles on spontaneous pneumorachis. All articles mentioned in PubMed, article bibliographies, and meeting abstracts until March 2021 were analyzed individually. Two hundred and two articles were identified, which were screened with inclusion criteria. Based on the selection criteria, 76 articles related to spontaneous pneumorachis were found, while 126 articles related to iatrogenic/traumatic pneumorachis were excluded. Of these, eight articles were written in a non-English language, two articles were inaccessible, and 18 articles on pediatric patients were excluded from the review. Forty-eight articles were included for final review and analysis, published over the past 40 years (1981-April 2021). The STrengthening, the Reporting of Observational studies in

Epidemiology (STROBE) diagram, mentions in detail the steps of screening and scrutinizing the various articles during the selection process of the articles for this review (Figure 5).

Selection and Inclusion Criteria

The literature search was done by two independent and trained researchers. Abstracts of all the articles were screened to ensure appropriateness. Articles included were cases involving humans and published in the English language only. We excluded the articles that included pediatric cases (18 cases) and published in a language other than English (8 cases). Hence, 48 articles detailing 49 cases published from 1994 till March 2021 were included in the final interpretation and discussion (Figure 5).

Data Extraction

All selected articles were thoroughly studied: data were extracted and entered in a predefined table in a Word version 16.17 (Microsoft Corp, Redmond, WA) spreadsheet. Details on demographic characteristics, clinical features, comorbidities, management (antibiotics, surgery, oxygen supplementation), need for mechanical ventilation, and the outcome was extracted from each article.

Results

Patient Characteristics

There were 49 cases in 48 case reports (the case report by Martins et al had 2 cases).⁵ There were eight females, 40 males, and the sex of one patient was unknown. There were 12 cases between the ages of 18 to 20, 13 cases between the ages of 21 to 30, 4 cases between the ages of 31 to 40, 18 cases beyond the age of 40, and the age of 2 cases were unknown. There were no comorbidities in 25 patients. Asthma and diabetes mellitus were the most common comorbidities present in 7 patients each.⁶ There were no comorbidities in 24 patients, the comorbidities of 4 patients were unknown, and adenocarcinoma of the colon was present in 2 patients. Depression, anorexia nervosa, bullous lung disease, multiple sclerosis, Pancoast tumor, and diverticulitis were present in 1 patient each (Table 1).

The trigger was unknown in 7 patients, upper respiratory tract infection/bronchitis/common cold in 6 cases, asthma exacerbation in 5 cases, emphysematous pyelonephritis in 3 patients, cocaine use in 3 patients, vomiting caused by diabetic ketoacidosis in 3 patients, an acute bout of dry cough in 2 cases and vacuum intervertebral disc in 2 cases.⁷⁻⁹ Other causes include Valsalva maneuver, choking, ketamine sniffing, strenuous exercise, weightlifting, entero dural fistula, mucous plugging, pneumocystis

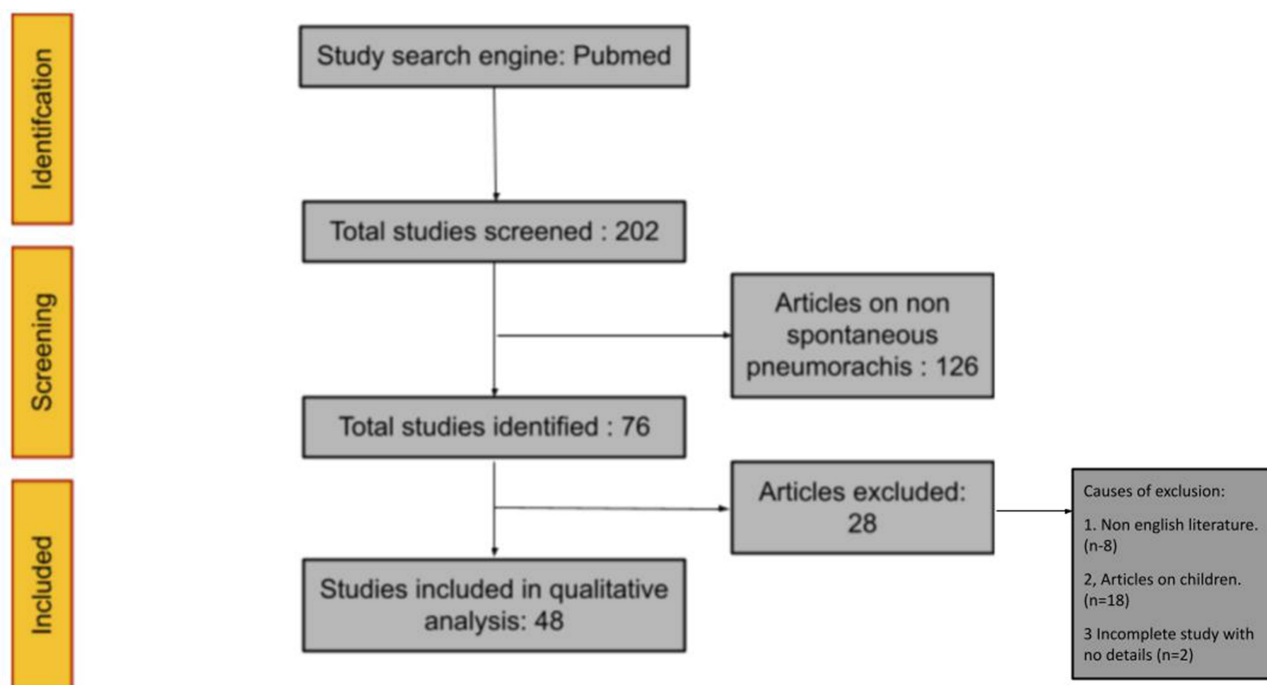


Figure 5 STROBE diagram depicting the selection process stepwise during the literature search for articles on nonspontaneous pneumorachis.

Table 1 Recent Cases of Spontaneous Pneumothorax and Management Reported in Literature

S. NO	Author	Age/Sex	Comorbidities	Acute Predisposition	Leaks in Spaces other than the Spinal Cord	Treatment	Intubation	Outcome
1	Navriya et al ⁽¹³⁾	65/m	Diabetes mellitus	Emphysema pyelonephritis	Kidney	DJ stenting	No	Discharged
2	Burns et al ⁽⁶²⁾	36/m	None	Cocaine insufflation followed by Valsalva maneuver	Pneumomediastinum, subcutaneous emphysema	Oxygen	No	Discharged
3	Bally et al ⁽⁶⁴⁾	Mid 20s/M	None	Choking	Subcutaneous emphysema, pneumomediastinum	Supportive	No	Discharged
4	Llewellyn et al ⁽⁷⁾	21/m	None	Upper respiratory tract infection	Subcutaneous emphysema, pneumomediastinum	Nasal cannula	No	Discharged
5	Williams et al ⁽⁸⁾	22/m	None	Ketamine sniffing	Subcutaneous emphysema, pneumomediastinum	Nonrebreather	No	Discharged
6	Niemann et al ⁽¹⁰⁾	19/m	None	Valsalva maneuver	Pneumothorax, pneumomediastinum, subcutaneous emphysema	Supportive	No	Discharged
7	Ramses Bedolla-Pulido et al ⁽⁸²⁾	18/m	Asthma	Acute exacerbation of asthma	Subcutaneous emphysema, pneumomediastinum	Srteroids, oxygen	No	Discharged
8	Liu et al ⁽⁶⁹⁾	18/m	None	Common cold	Subcutaneous emphysema, pneumomediastinum, pneumothorax	Conservatively	No	Discharged

(Continued)

Table 1 (Continued).

S. NO	Author	Age/Sex	Comorbidities	Acute Predisposition	Leaks in Spaces other than the Spinal Cord	Treatment	Intubation	Outcome
9	Radhika Nair et al ⁽⁷¹⁾	Unknown/f	Asthma	Asthma exacerbation	Pneumomediastinum, pneumoperitoneum, pneumothorax and subcutaneous emphysema	Symptomatic treatment	No	Recovered
10	Valiyakath et al ⁽⁸⁷⁾	21/f	None	Bronchitis	Pneumomediastinum, subcutaneous emphysema	Oxygen, intravenous antibiotics	No	Discharged
11.	Heckman et al ⁽⁸⁹⁾	20/m	None	Bronchitis.	Subcutaneous emphysema, prevertebral, parapharyngeal, carotid spaces, pneumomediastinum	Azithromycin, ceftriaxone, analgesia	No	Discharged
12	Ramasamy et al ⁽⁶⁶⁾	20/m	None	Strenuous exercise	Pneumomediastinum, subcutaneous emphysema	Nasal cannula oxygen, analgesics, antibiotics	No	Discharged
13	Gomez et al	60/m	Diabetes, ventral hernia	Emphysematous pyelonephritis	Kidney	Vasopressors, Broad spectrum antibiotics.	No	Discharged
14	Germino et al ⁽¹⁴⁾	58/m	None	Weightlifting	Craniocervical hyperpneumatization, pneumomediastinum, subcutaneous emphysema	Conservative management.	No	Discharged
15	Temrel et al ⁽⁶³⁾	28/m	None	Cocaine sniffing	Emphysema, pneumomediastinum	Conservative management	NO	Discharged
16.	Kanu et al ⁽⁷⁰⁾	19/m	None	Crack cocaine use	Subcutaneous emphysema, pneumomediastinum.	Conservative	No	Discharged

17	Schomig et al ⁽¹⁶⁾	67/m	Diverticulitis s/p clipping	Enterodural fistula caused by ingestion of dental prosthesis	Pneumocephalus	Lumbar puncture, vancomycin, ceftriaxone, Repair of entero dural fistula	No	Discharged
18	Mahajan et al ⁽¹⁷⁾	18/f	Asthma	Mucous plugging of the right main bronchus.	Pneumomediastinum, subcutaneous emphysema, right pneumothorax	Antibiotics, bronchodilators, bronchoscopic removal of mucous plugs.	Yes	Discharged
19	Patel et al ⁽⁵⁰⁾	20/m	None	Upper respiratory tract infection	Subcutaneous emphysema, pneumomediastinum, pneumothorax.	Left side chest tube, oxygen through NRB.	No	Discharged
20	Eroglu et al ⁽⁶⁰⁾	44/F	None	Unknown	None	Anti inflammatory	No	Discharged
21	Saleem et al ⁽¹⁸⁾	28/m	None	Pneumocystis pneumonia with new onset AIDS	Subcutaneous emphysema, pneumomediastinum.	Oxygen through non rebreather, Trimethoprim-Sulfamethoxazole, Prednisone, Bronchoscopy for diagnosis.	No	Discharged
22	Kirkham et al ⁽¹⁾	21/m	Asthma	Asthma exacerbation	Subcutaneous emphysema, pneumopericardium	Observation	No	Discharged
23	Ozkan et al ⁽³⁾	31/m	Bullous lung disease	Spontaneous rupture of bulla	Pneumothorax, subcutaneous emphysema	Chest tube, bulla removal surgery	No	Discharged
24	Kazimirko et al ⁽²⁹⁾	53/m	Right superior sulcus lung tumor with adjacent vertebral invasion with pathologic fracture of T1.	Bronchopleurodural subarachnoid fistula	Pneumocephalus	C-collar and antibiotics.	No	Discharged but died 3 months later from cancer progression
25	Krishnan et al ⁽²⁰⁾	30/M	Unknown	Unknown	Subcutaneous emphysema, pneumomediastinum	Subcutaneous incisions, symptomatic medications	No	Discharged

(Continued)

Table 1 (Continued).

S. NO	Author	Age/Sex	Comorbidities	Acute Predisposition	Leaks in Spaces other than the Spinal Cord	Treatment	Intubation	Outcome
26	Jensen et al ⁽⁹⁾	Unknown	Anorexia nervosa	Bouts of self induced vomiting	Unknown	Conservative medical approach and treatment of anorexia nervosa	Unknown	Unknown
27	Moayedi et al ⁽²⁸⁾	76/f	Multiple sclerosis, diabetes mellitus	Sacral decubitus ulcer	Pneumocephalus, subcutaneous emphysema	IV antibiotics - cefepime, metronidazole; levetiracetam	Yes	Passed away 6 days later from sepsis after being made comfort measures
28	Martins et al ⁽⁵⁾	20/m, 22/m	None	Acute bout of dry cough.	Pneumomediastinum, subcutaneous emphysema	Bedrest, oxygen, and opioids for cough control	No	Discharged
29	Liao et al ⁽⁶⁸⁾	19/m	None	Strenuous exercise	Subcutaneous emphysema, pneumomediastinum	Conservative treatment	No	Discharged
30	Eisa et al ⁽⁶⁴⁾	18/m	None	Upper respiratory tract infection	Subcutaneous emphysema, pneumothorax, pneumomediastinum	Oxygen, conservative management	No	Discharged
31	Amara et al ⁽⁸¹⁾	21/m	None	Fever and vomiting in the setting of meningitis	Pneumomediastinum, subcutaneous emphysema	Ceftriaxone, lumbar puncture, bronchoscopy, laryngoscopy	Yes	Died from sepsis
32	Iacoangeli et al ⁽²¹⁾	52/F	Adenocarcinoma of colon	Transsacral cerebrospinal fluid leak	Pneumocephalus	Lumbosacral laminectomy and duraplasty; tumor removal and omental covering of the pelvis	No	Discharged home
33	Tafreshi et al ⁽²²⁾	66/m	Adenocarcinoma of colon T3 with history of abdominoperineal resection and recurrence with presacral mass.	Enterocutaneous fistula from left natal cleft	None	Pelvic exenteration	No	Discharged.
34	Lee et al ⁽²³⁾	76/m	Unknown	Vacuum intervertebral disc	None	Left partial hemilaminectomy	No	Discharged

35	Kumaran et al ⁽²⁴⁾	51/m	None	Grade I anterolisthesis of L5 over S1	None	None	Spondylolisthesis correction surgery	No	Unknown
36	Amit et al ⁽²⁵⁾	60/m	Unknown	Perforated sigmoid colon	Pneumocephalus	Hartmann's procedure; broad-spectrum intrathecal antibiotics	No	Resolution of pneumocranium and pneumorachis	
37	Al-Mufarrej et al ⁽⁸⁸⁾	20/m	None	Unknown	Pneumoperitoneum, pneumomediastinum, subcutaneous emphysema	Self-limited	No	Resolved	
38	Song et al ⁽⁸²⁾	18/m	None	Unknown	Pneumomediastinum, subcutaneous emphysema	Dextromethorphan 45 mg daily	No	Discharged	
39	Kim et al ⁽¹²⁾	64/m	Unknown	Vacuum disc L5-S1.	None	Conservative	No	Resolved	
40	Drolet et al ⁽³⁶⁾	18/m	Type I diabetes mellitus	Diabetic ketoacidosis	Pneumomediastinum, subcutaneous emphysema	Observation	No	Discharged	
41	Eesa et al ⁽³⁸⁾	18/m	Asthma	Asthma exacerbation	Pneumomediastinum, subcutaneous emphysema	Unknown	Unknown	Unknown	
42	Oertel et al ⁽⁴⁾	19/m	Asthma, diabetes mellitus	Diabetic ketoacidosis	Pneumomediastinum, pneumoperitoneum, subcutaneous emphysema, pneumocephalus	Otolaryngological exploration	No	Discharged	
43	Eesa et al ⁽³⁸⁾	18/m	Asthma	Asthma exacerbation	Pneumomediastinum, subcutaneous emphysema, retropharyngeal space	Oxygen therapy, bronchodilators, and antibiotics	No	Discharged	
44	Yamamoto et al ⁽¹⁵⁾	63/m	Depression, Diabetes mellitus	Emphysematous pyelonephritis	Right iliopectus muscle, kidney	Intravenous vancomycin 1 g BID for 1 week and meropenem 1 g TID for 4 weeks	No	Discharged	

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Table 1 (Continued).

S. NO	Author	Age/Sex	Comorbidities	Acute Predisposition	Leaks in Spaces other than the Spinal Cord	Treatment	Intubation	Outcome
45	Pangtey et al ⁽⁷²⁾	35/m	None	Unknown	Pneumothorax, pneumomediastinum, subcutaneous emphysema	Chest tube	No	Discharged
46	Ripley et al ⁽⁶⁾	23/m	Type I diabetes mellitus	Diabetic ketoacidosis	Pneumomediastinum, subcutaneous emphysema	Standard treatment for DKA and supplemental oxygen	No	Discharged
47	Fonseca et al ⁽¹⁹⁾	20/F	None	Unknown	Subcutaneous emphysema, pneumomediastinum	Bronchoscopy, conservative management	No	Discharged
48	Song et al ⁽²⁾	72/F	None	Unknown	None	C7 total laminectomy	No	Discharged

pneumonia, spontaneous rupture of the bulla, bronchopleural dura subarachnoid fistula, bouts of self-induced vomiting in anorexia nervosa, vomiting induced by meningitis, transsacral cerebrospinal fluid leak, enterocutaneous fistula from the left natal cleft, and anterolisthesis of L5 over S1 in 1 patient each.¹⁰⁻¹²

Other Sites of Involvement

The spinal cord was the only site of air accumulation in 5 cases. Concomitant pneumomediastinum was reported in 30 cases, subcutaneous emphysema was reported in 33 cases, pneumothorax in 8 cases, pneumocephalus in 6 cases, kidney in 3 cases, deep spaces of the neck in 2 cases, and right iliopsoas muscle in 1 case (Figure 6).

Management Strategies

The management was supportive in most of the cases, except for a few cases. The supportive treatment included a combination of oxygen supplementation, analgesics, and cough suppressants. Only three patients required intubation and mechanical ventilation. It was unknown whether an intubation was required in 2 patients. Patients with asthma exacerbation were treated with steroids and nebulizers. Navriya et al report that their patient with emphysematous pyelonephritis was managed with double J stenting.¹³ The patient in Gomez et al's case report required vasopressor and broad-spectrum antibiotics for emphysematous pyelonephritis, as did the patient reported by Yamamoto et al^{14,15} Schomig et al had to do a lumbar puncture followed by repair of enterodural fistula for the treatment of pneumorachis caused by enterodural fistula.¹⁶ Mahajan et al's patient required bronchoscopic removal of mucous plugging, while another patient of Saleem et al required bronchoscopy to diagnose pneumocystis pneumonia.^{17,18} Fonseca et al also performed a bronchoscopy, although the reason for the procedure is unclear.¹⁹ A total of 3 patients required chest tubes for the concomitant pneumothorax, following which pneumorachis resolved. Krishnan et al performed subcutaneous incisions to help relieve the trapped mediastinal air, subsequently leading to the resolution of pneumomediastinum.²⁰ Lumbosacral laminectomy with duraplasty was employed by Iacoangeli et al for management of pneumorachis secondary to transsacral cerebrospinal fluid leak caused by adenocarcinoma of the colon.²¹ C7 total laminectomy (Song et al),⁸² otolaryngological exploration (Oertel et al),⁷⁷ Hartman's procedure (Amit et al), spondylolisthesis correction surgery (Kumaran et al), left partial hemilaminectomy (Lee et al), and pelvic exenteration (Tafreshi et al) were some of the other

Other sites of involvement

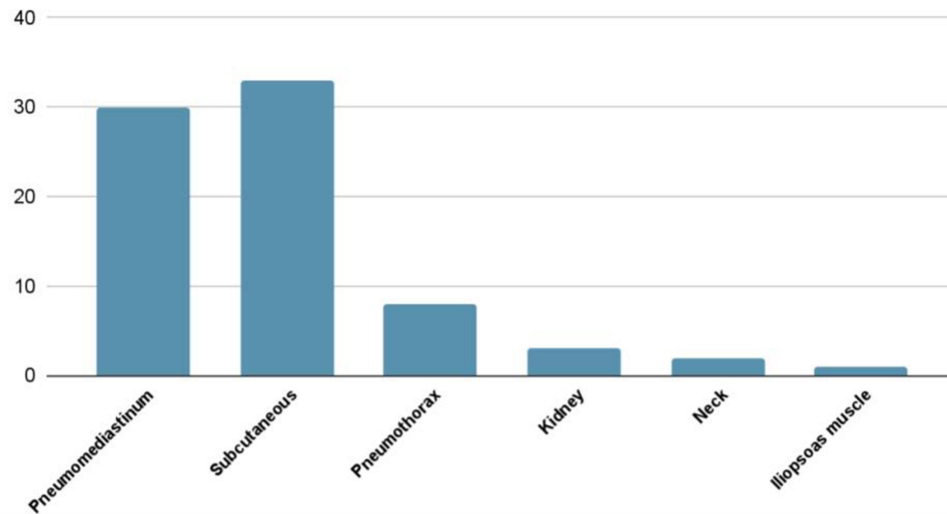


Figure 6 Various sites of air leaks in addition to pneumorachis in our study cohort.

procedures employed by authors for management of the inciting factor for the pneumorachis.^{22–27}

Outcome

The outcome was unknown in 2 cases, one patient died of sepsis, and one patient died three months after being discharged due to cancer progression.^{28,29} The outcome was positive in 45 out of the 49 cases, with the patients having either resolution of the pneumorachis or being discharged home.

Discussion

Pneumorachis can be classified based on etiology as traumatic, iatrogenic, or spontaneous.³² A 2010 systematic review of pneumorachis identified trauma to the respiratory tract as the causative factor in 52% of cases. Traumatic injury to the skull (especially skull base and sinus fractures) was the second most common cause.³³ Iatrogenic causes include lumbar puncture, epidural analgesia, spinal surgeries, and abdominal surgeries.^{34,35} Epidural abscesses due to anaerobic pathogens can cause the air in the epidural space, especially in patients who have diabetes or Crohn's disease.³⁶ Intra-abdominal infections (especially emphysematous pyelonephritis) with anaerobic/facultative anaerobic bacteria such as *Citrobacter koseri* can also generate pneumoperitoneum with subsequent invasion of air into the spinal canal.¹⁵ Spontaneous pneumorachis can occur as a result of respiratory conditions and complications that incite barotrauma.^{37,38} Asthma is a common, chronic respiratory disease affecting

8% of adults in the U.S.A, and some of the rare but potentially life-threatening complications of an asthma exacerbation include pneumothorax and pneumomediastinum pneumopericardium, pneumorachis, tracheoesophageal fistula, and anoxic brain injury.^{39–41} Asthma was identified as the most common trigger for spontaneous pneumorachis in our review.⁴² Rarely, spontaneous extradural pneumorachis can develop as a complication of severe degenerative disc disease. Intervertebral disc vacuum phenomenon is a finding present in 1–2% of spinal radiographs and has an estimated prevalence of 20% in elderly population.⁹³ Enlargement of this clefts in the intervertebral disc can cause a negative pressure gradient pulling gases into the space. The disk fragment containing gas could herniate into the spinal canal or the gas could get expelled into the epidural space through a weak spot in the annulus fibrosis, occasionally causing clinical presentation akin to a herniated disc.

Clinical Anatomy

Due to continuity between the cranial cavity and spinal canal, it is expected that cranial injuries communicating with the external atmosphere could cause pneumocephalus and consequent pneumorachis. However, as mentioned above, respiratory tract injuries account for most cases of pneumorachis. Defining the relations between the respiratory tract, mediastinum, and vertebral column is key to understanding the mechanism of spontaneous pneumorachis.

The mediastinum is the space within the thoracic cavity bordered superiorly by the thoracic inlet, inferiorly by the diaphragm, and excludes the lungs and pleural cavities. Within the mediastinum, the air is usually seen in the trachea, bronchi, bronchioles, alveoli, and esophagus, all of which are a potential source for pneumomediastinum and pneumorachis. Being the thinnest portion within the respiratory tree, alveolar rupture under conditions of the high trans alveolar pressure gradient is possibly the most common source of free air. If the rupture involves the visceral pleura, the patient will develop a pneumothorax. The air could also escape into the pulmonary interstitium from where it tracks along the bronchovascular bundle to reach the hilum from where it escapes into the mediastinum (Macklin effect).⁴³ From here, air can dissect into the posterior mediastinum and separates the parietal pleura from the vertebral column gaining access into the epidural space via the intervertebral foramen.⁴⁴ In the absence of any fascial barriers between the posterior mediastinum and epidural space, air can freely communicate between these compartments.⁴⁵

There is no direct communication between the mediastinum and the pericardial cavity. Pneumopericardium typically occurs due to macro perforation of the pericardium and communication with the respiratory or GI tract in the setting of trauma, pneumothorax, volutrauma due to mechanical ventilation, fistulas in the setting of malignancies, etc.⁴⁶ Laparoscopic surgeries could occasionally cause pneumopericardium. This is attributed to the presence of congenital patent pathways between pleura, pericardium, and peritoneal cavities. Our patient did not have a pneumothorax or pneumoperitoneum; hence, it is likely that the large volume of air was able to push into the pericardial space near the pulmonary vein Ostia where the parietal pericardium reflects onto the visceral pericardium creating the weakest spot.⁴³

Pathophysiology

Literature review revealed one case of pneumorachis associated with cannabis use which was attributed to using a homemade bong with flow restricted intake, which forced the patient to inhale forcibly against high resistance, in turn creating a high negative intrathoracic pressure inducing barotrauma.^{47,48} This phenomenon is called Müller's maneuver and can be considered the reverse of the Valsalva maneuver. A similar mechanism could explain

pneumorachis and pneumomediastinum in our patient too. In this case, instead of an external resistance to airflow, bronchospasm induced by the cannabis smoke would have forced the patient to inhale against a tight airway generating high negative intrapleural pressures, while coughing against the resistance would have generated high positive intra-alveolar pressures, thereby increasing trans alveolar pressure gradient both during inspiration and expiration which resulted in the barotrauma. The chances of a pneumomediastinum causing pneumorachis increase with the amount of leaked air and acuity of onset.⁴⁹

Clinical Presentation

Spontaneous pneumorachis in the absence of intraspinal causes like epidural abscesses are associated with pneumomediastinum or subcutaneous emphysema.⁵⁰ Asthma exacerbation is the most common triggering factor estimated to be present in 20–30% of all cases of spontaneous pneumomediastinum in children and adolescents.⁵¹ Other predisposing conditions include respiratory tract infections, activities inducing Valsalva maneuver, activities causing Müller's maneuver, severe vomiting, esophageal rupture, foreign body aspiration, vaping and inhalational drug use.^{52–63} The presence of these predisposing factors along with signs and symptoms should raise the suspicion for spontaneous pneumomediastinum (SPM) and possible pneumorachis.⁶⁴ SPM is more common among children and adolescents compared to older age groups. Males are more likely to be affected, and so does a tall, lean body habitus.^{65,66}

The most common presenting complaint is retrosternal, pleuritic chest pain radiating to the neck or shoulders of acute onset after an inciting event.^{67,68} Even when no triggering factor is present, SPM should be considered in the differential diagnosis of any young adult presenting with acute onset chest pain.^{69,70} Other common symptoms include subcutaneous emphysema with crepitus, especially in the neck, neck pain, dyspnea, cough, odynophagia, and dysphagia. Esophageal perforation (Boerhaave syndrome) could also present with a similar presentation and should be considered, especially in patients who have severe vomiting; but they typically present with signs and symptoms of shock.

Subcutaneous emphysema detected over the neck or precordium is an extremely sensitive and specific sign of SPM.^{71,72} Hamman sign, a precordial crunching sound

synchronous with systole, especially in the left lateral decubitus position, may be present in up to 18% of patients.⁷³ Findings such as hypotension, severe respiratory distress, unilaterally diminished breath sounds, and distended neck veins should raise the suspicion for more serious etiologies such as tension pneumomediastinum, pneumothorax, or esophageal perforation.

Neurological Signs and Symptoms

Pneumorrhachis is asymptomatic in most cases but can cause neurologic symptoms, including spinal cord compression in about 10% of patients.^{74,75} As pneumorrhachis by itself rarely causes spinal cord compression, we should search for alternative explanations such as epidural hematomas, abscesses, herniation, etc, if neurological symptoms are present.^{76–81}

Diagnosis

Pneumorrhachis is almost always found in combination with air in other cavities such as pneumothorax, pneumomediastinum, or subcutaneous emphysema.⁸² Ultrasound is gaining popularity as the imaging of choice in the emergency department for the fast and accurate detection of pneumomediastinum, but it is not useful for detecting pneumorrhachis.^{83,84} Pneumorrhachis can be diagnosed on spinal X-ray.⁸⁵ A CT scan is the diagnostic test of choice.^{86,87} An MRI is not required unless signs of cord compression are present and other etiologies such as contusion, hematoma, herniation, or compressing bone fragments need to be ruled out. The characteristic radiological features of SPM and pneumorrhachis, as demonstrated in our patient, are as below:

Treatment

There are no guidelines for the management of pneumorrhachis.⁸⁸ Serial neurological examinations are recommended, while serial imaging for pneumorrhachis is not required unless there is a concern for cord compression.⁸⁹ The development of neurological symptoms warrants surgical decompression; otherwise, the management is conservative.⁴ Treatment of the inciting etiology, such as bronchodilators and oxygen supplementation for asthma exacerbation, abscess drainage, chest tube for pneumothorax, etc. are required.

Clinical Outcomes

Clinical outcomes are related to the etiology of pneumorrhachis. The one patient who died during the acute course of illness in our review had sepsis. Radiological resolution

would occur in most cases with conservative management. Treatment of the cause will curtail ongoing air leaks allowing the intra-spinal air to get reabsorbed within hours to days. Residual neurological deficits are rare.³³

Conclusion

Clinicians should be aware of the possibility of pneumorrhachis in the setting of acute asthma exacerbation. Although asymptomatic in most cases, it can rarely lead to serious complications such as spinal cord compression. Addressing the underlying etiology is more important than managing the pneumorrhachis per se. Treatment is conservative except in symptomatic cases where intervention is required.

Disclosure

The authors report no conflicts of interest in this work.

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