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CASE REPORT

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Pneumorrhachis secondary to exacerbation of asthma: A case report and literature review

Chris Zi-Fan Zhao[1] | Nadia Poci[1] | Daniel Niewodowski[1] | Amy Baker[2] | Christine McDonald[1,3,4]

[1]Department of Respiratory and Sleep Medicine, Austin Health, Heidelberg, Victoria, Australia

[2]Department of Radiology, Austin Health, Heidelberg, Victoria, Australia

[3]Institute for Breathing and Sleep, Heidelberg, Victoria, Australia

[4]Department of Medicine, University of Melbourne, Parkville, Victoria, Australia

Correspondence
Chris Zi-Fan Zhao, Department of Respiratory and Sleep Medicine, Austin Health, 145 Studley Rd, Heidelberg, Melbourne, VIC 3084, Australia.
Email: czfzhao@gmail.com

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Abstract

Pneumorrhachis is defined by the presence of air within the spinal cord. Spontaneous pneumorrhachis secondary to exacerbation of asthma is rare, and its management is rarely discussed. We present a case of spontaneous pneumorrhachis in the context of a viral exacerbation of asthma, followed by a systematic literature review of all available cases of pneumorrhachis in asthma exacerbation. A total of 25 case studies reported pneumorrhachis in 28 asthma patients, all of whom presented with concomitant pneumomediastinum. Investigation and exclusion for other potential aetiologies of pneumorrhachis such as trauma or infection occurred to varying extents and may depend on clinical presentation and degree of suspicion. No other contributing aetiologies were demonstrated in this review, and no patients required specific intervention for pneumorrhachis. Whilst pneumorrhachis is generally benign, management should revolve around standard care of asthma exacerbation, attention to potentially life-threatening differential diagnoses, and supportive care.

KEYWORDS

asthma, epidural emphysema, pneumomediastinum, pneumorrhachis

INTRODUCTION

Pneumorrhachis is defined by the presence of air within the spinal canal. It is an exceptional finding most often encountered in traumatic presentations or iatrogenic following procedures involving the spinal canal.[1] In the absence of trauma or iatrogenic causes, spontaneous pneumorrhachis has been reported secondary to respiratory tract infection, abdominal infections, asthma, vomiting, acute cough and is usually preceded by pneumomediastinum.[2]

There are no published guidelines for the management of pneumorrhachis[3] but it is generally considered benign and self-limiting.[2,4] Reports of exacerbation of asthma as a cause for spontaneous pneumorrhachis are few, and its management is rarely discussed.

We present a case of spontaneous pneumorrhachis in the context of a viral exacerbation of asthma in a young male. This is followed by a literature review obtained via a structured search of MEDLINE and PubMed of all available cases of pneumorrhachis in asthmatic patients and a discussion on management.

CASE REPORT

A 25-year-old never-smoking office worker with known severe eosinophilic asthma, presented to the emergency department following 2 days of progressive coryzal symptoms, dry cough, and dyspnoea. He experienced an acute onset of pleuritic chest and neck pain preceded by an episode of coughing. He reported no recent trauma or procedures prior to the presentation. He reported no cannabis smoking, no inhaled recreational drug use or any occupational exposures to inhalants.

The patient's asthma was managed with twice-daily high-dose inhaled-corticosteroid and long-acting beta-agonist and monthly mepolizumab injections, with good compliance. Prior to his acute illness, he used a salbutamol inhaler on average five times each day, though had not

required oral corticosteroid therapy in the antecedent 12 months. He has never required invasive ventilation or admission into intensive care and his most recent spirometry and TLCO were within normal limits.

On presentation, he was found to be tachycardic, tachypnoeic and mildly hypoxaemic requiring 2 L of supplemental oxygen via nasal cannulae to maintain oxygen saturation of 96%. He had a widespread polyphonic expiratory wheeze, and palpable subcutaneous emphysema across the chest. He presented with no neurological signs or symptoms and was afebrile. His initial pathology results demonstrated mild neutrophilia, an eosinophil count of 0.0, and a CRP of 23.9 mg/L though these were obtained following administration of intravenous dexamethasone pre-hospital. His initial venous blood gas demonstrated a mixed respiratory and metabolic acidosis with a lactate of 1.9 mmol/L. Testing for COVID-19 was negative, with positive nasal swab viral polymerase chain reaction testing for rhinovirus/enterovirus species. Plain chest radiography revealed extensive pneumomediastinum as well as chest wall and neck subcutaneous emphysema.

He was commenced on intravenous corticosteroid therapy, intravenous magnesium sulfate, and regular short-acting bronchodilators. Empirical oral antibiotic therapy was commenced for possible community-acquired pneumonia or possible mediastinitis causing pneumomediastinum. Computed tomography (CT) of the chest and neck performed to assess for possible oesophageal rupture confirmed the presence of large volume pneumomediastinum extending throughout the soft tissues of the neck and upper chest wall, and pneumorrhachis within the lower cervical spinal canal (Figure 1). No injury to the oesophagus or throat was demonstrated, and there was no pneumothorax.

This patient's pneumorrhachis and pneumomediastinum were managed conservatively. He responded well to routine management of asthma exacerbation and was weaned off oxygen over 48 h. During hospitalization he was monitored closely with serial neurological examinations and chest radiographs to ensure stability of his pneumomediastinum. He was discharged 3 days post-admission to complete a short course of oral corticosteroids and antibiotics and at follow-up, 4 weeks after discharge, he had only mild ongoing respiratory symptoms. Notably, 1 year following this patient's initial presentation, he represented with recurrent spontaneous pneumomediastinum in the setting of asthma exacerbation but without pneumorrhachis.

LITERATURE REVIEW

Methods

A structured search of MEDLINE and PubMed using the search terms 'pneumorrhachis/pneumorachis', 'epidural emphysema', 'intraspinal pneumocoele', 'spinal pneumatosis' 'aerorachia', 'pneumosaccus', 'air myelogram' and 'asthma' was performed on the 30th of July 2023. This search resulted in 33 articles in English which were assessed in their entirety by one author. This identified 19 case studies reporting pneumorrhachis in asthma patients which were included. A further six relevant case studies were referenced by articles of the initial search and included. Articles reporting patients of all ages were accepted. Information relating to age, gender, clinical presentation, examination, investigation and imaging findings, management and clinical course were collected.

Results

A total of 25 case studies reported 28 asthma patients presenting with pneumorrhachis (Table 1). Patient's age ranged

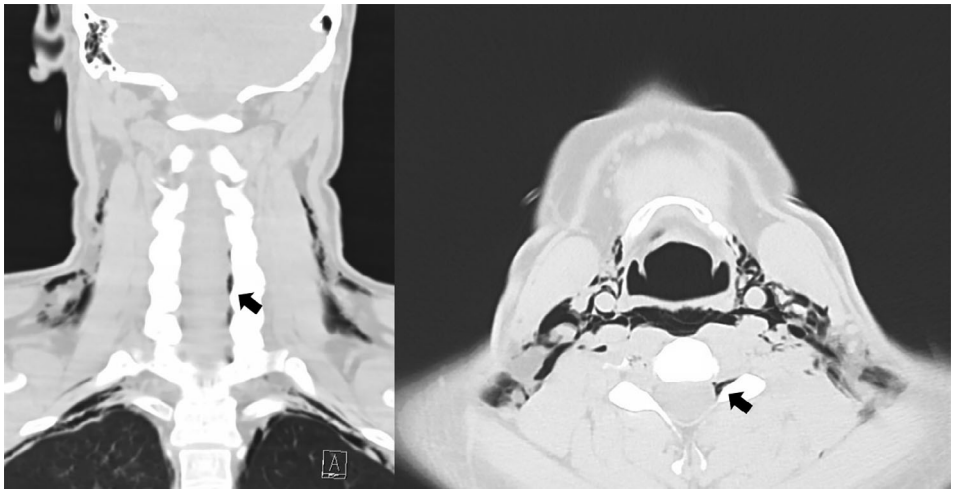

FIGURE 1 Coronal and axial views of gas within the extradural spinal canal (pneumorrhachis) at the lower cervical levels (arrows). The axial image demonstrates the path of air dissection through the intervertebral foramen into the epidural space. There is extensive air extending in the soft tissues of the neck and upper anterior chest wall.

TABLE 1 Clinical presentation and management of all available reports of asthma patients presenting with pneumorrhachis.

Pt	First author, journal	Year; volume: Page	Age	Gender	Presentation	PM	PT	Management
1	Tsuji H, J Comput Assist Tomogr	1989;13:38–9	18	M	Dyspnoea, wheeze	Yes	–	Bronchodilators, steroids, antibiotics
2	Drevelengas A, Eur J Radiol.	1994;18:122–3	–	M	Dyspnoea, cough, chest pain, hypoxia	Yes	No	ICU
3	Kakitsubata Y, Acta Radiol.	1994;35:305–6	–	M	Wheeze, cough, neck pain	Yes	–	Bed rest
4	Caramella D, Pediatr Radiol.	1997;27:929–31	13	M	Dyspnoea, wheeze, cough, fever, chest pain, hypoxia, neck swelling	Yes	No	Bronchodilators, steroids, antibiotics, CT-Brain
5	Caramella D, Pediatr Radiol.	1997;27:929–31	7	–	Asthma exacerbation	Yes	–	–
6	Pifferi M, Pediatr Pulmonol.	1997;24:125–6	13	M	Cough	Yes	–	–
7	Pifferi M, Pediatr Pulmonol.	1997;24:125–6	7	M	Dyspnoea, wheeze, cough, hypoxia	Yes	–	–
8	Van der Klooster JM, Neth J Med.	1998;52:150–4	26	M	Dyspnoea, wheeze, cough, fever, neck and chest pain	Yes	No	Bronchodilators, steroids, antibiotics, oxygen
9	Dosios T, Euro J Cardiothoracic Surg.	2000;18:123	15	M	Asthma exacerbation	Yes	No	–
10	Oertel MF, J Neurol Neurosurg Psychiatry.	2005;76:1036	19	M	Cough, fever, nausea and vomiting	Yes	No	Antibiotics, antitussives, surgical review
11	Eesa M, Acta Radiol.	2006;47:672–4	18	M	Wheeze, no cough, neck swelling	Yes	–	Bronchodilators, antibiotics, oxygen, ICU
12	Manden PK, Ann Thorac Med.	2009;4:143–5	17	M	Dyspnoea, wheeze, cough, fever, hypoxia	Yes	No	Bronchodilators, steroids, antibiotics, oxygen
13	Bhadane S, Med J Armed Forces India.	2011;67:93	14	M	Dyspnoea, wheeze, cough	Yes	No	Bronchodilators, steroids, antibiotics, oxygen
14	Karaoglan A, Turk Neurosurg.	2011;21:666–8	8	F	Respiratory distress, cough, fever, hypoxia, neck swelling	Yes	No	Bronchodilators, antibiotics, oxygen
15	Hanada T, Respir Investig.	2012;50:62–5	13	M	Wheeze, cough, fever, neck swelling	Yes	No	Inhaled corticosteroids
16	Hanada T, Respir Investig.	2012;50:62–5	15	M	Wheeze, cough, fever	Yes	No	Bronchodilators, steroids, MRI
17	Adachi T, Respiration.	2012;84:69	22	M	Dyspnoea, wheeze, cough, chest pain, hypoxia	Yes	No	Bronchodilators, steroids, oxygen
18	Girard C, Am J Respir Critical Care Med.	2014;189:e69	18	F	Wheeze, facial and neck swelling, nasal voice	Yes	No	Bronchodilators, steroids
19	Mahajan PS, Int Med Case Rep J.	2014;7:35–9	18	F	Dyspnoea, cough, chest pain	Yes	Minimal	Bronchodilators, antibiotics, oxygen, bronchoscopy
20	Gentili A, Minerva Anestesiol.	2014;80:508–9	9	[a]	Dyspnoea, wheeze, cough, back pain, hypoxia	Yes	Yes	Bronchodilators, steroids, antibiotics, oxygen, ICU, NIV, bronchoscopy
21	Teixeira C, Prensa Med Argent	2015;101:405–8	18	F	Dyspnoea, wheeze, cough, hoarse voice fever, neck pain, neck swelling, hypoxia, fever, odynophagia, hoarse voice	Yes	–	Steroids, antibiotics, oxygen
22	Kirkham J, J Osteopath M	2016;116:119	21	M	Dyspnoea, wheeze, cough, chest pain	Yes	–	–
23	Hochhegger B, Radiol Bras.	2018;51:268	5	M	Dyspnoea, wheeze, cough, fever, nausea and vomiting	Yes	No	Bronchodilators, steroids, antibiotics, oxygen
24	Radhika Nair K, J Assoc Physicians India.	2018;66:70–2	18	F	Dypsnoea, wheeze, cough, chest pain, facial and neck swelling	Yes	Minimal	Bronchodilators, antibiotics, oxygen
25	Gutiérrez-Morales I, Arch Bronconeumol.	2019;55:588	20	M	Asthma exacerbation	Yes	No	–

(Continues)

TABLE 1 (Continued)

Pt	First author, journal	Year; volume: Page	Age	Gender	Presentation	PM	PT	Management
26	Ramses Bedolla-Pulido T, J Asthma.	2019;56 (12):1356–9	18	M	Dypsnoea, cough, chest pain, facial/neck swelling	Yes	No	Bronchodilators, steroids, antibiotics
27	Yilmaz F, Acta Biomed.	2021;92: e2021141	19	M	Wheeze, cough, neck and chest pain	Yes	No	Bronchodilators, steroids, oxygen, surgical review
28	Alampoondi Venkataramanan SV, J Asthma Allergy.	2021;14:1539–54	21	M	Dyspnoea, wheeze, cough, sore throat, neck pain, neck swelling	Yes	Yes	Bronchodilators, steroids, oxygen

Abbreviations: CT, computed tomography; ICU, intensive care unit; MRI, magnetic resonance imaging; NIV, non-invasive ventilation; PM, pneumomediastinum; PT, pneumothorax; –, Not specified.

between 5 and 26 years, and approximately 75% of the patients were male. All cases presented with respiratory symptoms including dyspnoea, wheeze, or cough. Pain or subjective sensation of swelling in the chest, neck or face was present in 17 patients. Nine patients presented with fever, and at least eight patients were found to be hypoxaemic on presentation. No patients presented with neurological symptoms or signs. Pneumomediastinum on plain chest radiography or CT was found in all patients. Three patients were found to have concomitant pneumothoraces which did not require intervention.

Patients were generally managed with a combination of bronchodilators and intravenous corticosteroids. Fourteen patients were managed with antibiotics. Thirteen patients were managed with oxygen therapy, in the absence of hypoxaemia in some cases.

No patients required specific invasive intervention for pneumorrhachis. One patient was reviewed by the surgical team for exclusion of nasopharyngeal abscess, another underwent magnetic resonance imaging of the spine for exclusion of cord compression, and one underwent a CT of the brain for exclusion of pneumocephalus. Two patients underwent bronchoscopy; one for exclusion of airway trauma as an aetiology for pneumomediastinum, the other as intervention for mucus plugging. No investigations yielded results requiring further management. Whilst infrequently reported, time of inpatient stay ranged between 1 and 12 days. Four patients were followed up with repeat CT imaging and all demonstrated resolution of pneumomediastinum and pneumorrhachis between 1 and 3 weeks.

DISCUSSION

Pneumorrhachis is infrequently reported in the literature and most often in isolated case studies.[3] The aetiology of pneumorrhachis in asthma is thought to be as a result of extension of pneumomediastinum into the epidural space via intervertebral foraminae.[5] Pneumomediastinum in asthma is itself presumed to occur as a result of alveolar rupture and dissection of air along broncho-vascular sheaths, known as the Macklin effect.[6] In this review, all patients with pneumorrhachis in the setting of asthma exacerbation had concomitant pneumomediastinum.

Given all patients in this case series were asymptomatic of their pneumorrhachis, care delivered to patients primarily involved asthma exacerbation standard of care. Fever was reasonably common, mostly without subsequent confirmed infective focus. Antibiotics were prescribed in more than half of patients, often in the setting of haemodynamic instability or as empiric treatment for potential mediastinitis. Epidural infection is a possible serious differential of pneumorrhachis, however, no case studies reported this as an indication for antibiotic therapy.

Investigation and exclusion of potential traumatic aetiologies of pneumorrhachis, such as tracheobronchial or oesophageal injury,[7] may depend on clinical presentation and degree of suspicion. Cross-sectional imaging of the chest

may be sufficient to exclude this, although in some cases further investigations were pursued such as bronchoscopy or oral contrast studies.[8,9] No included studies demonstrated structural injury in their further investigations. Whilst there have been isolated reports of pneumorrhachis causing neurological sequelae requiring surgery,[10] no patients in this series presented with neurological signs or symptoms.

High-concentration oxygen therapy may be considered, as it may promote re-absorption of gas into soft tissue in subcutaneous emphysema, pneumomediastinum, and presumably pneumorrhachis.[11] Anti-tussive medication may also be considered if cough is felt to be a significant precipitating factor in pneumomediastinum.[12]

In conclusion, pneumorrharchis in asthma is rare and most commonly occurs as a sequela of significant pneumomediastinum. Whilst pneumorrhachis is generally benign, management should revolve around standard care of asthma exacerbation, attention to potentially life-threatening differential diagnoses, and supportive care. Consideration for investigation of structural injuries contributing to pneumorrhachis depends on clinical suspicion, though no injuries were demonstrated in this case series. The use of empiric antibiotic therapy and supplemental oxygen therapy is common.

AUTHOR CONTRIBUTIONS

Dr Chris Zhao contributed to conceptualisation, literature review, preparation of initial manuscript and revision of final manuscript. Dr Nadia Poci, Dr Daniel Niewodowski and Prof. Christine McDonald contributed to conceptualisation and review of final manuscript. Dr Amy Baker contributed to review of imaging and review of final manuscript.

CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

ORCID

Chris Zi-Fan Zhao https://orcid.org/0000-0002-9223-6293

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