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Case report

Simultaneous *Streptococcus pneumoniae* empyema in fraternal twins

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ABSTRACT

Streptococcus pneumoniae is the most common bacterial cause of community acquired pneumonia. The current trend in *Streptococcus pneumoniae* infections has been the rise of multi-drug resistance in the last two decades. We present the case of a pair of 16-month old African-American fraternal twins who presented to the emergency room on the same day for symptoms consistent with pneumonia. Upon further examination, the twins showed remarkably similar symptoms, and cultures revealed penicillin-resistant *Streptococcus pneumoniae* in both twins. The pneumonia affected both twins in the same way, but a tomography scan did not reveal any shared anatomical abnormalities to account for this near-identical progression. In a review of literature and case reports, there are no reported cases of fraternal twins with simultaneous or non-simultaneous pneumococcal pneumonia or effusions. This case suggests that there may be possible anatomical abnormalities in the fraternal twins which were not evident in routine testing that may have led to near-identical illnesses. The pathophysiology of the simultaneous and near identical infections is not clear but may reflect subtle genetic factors in the siblings.

Introduction

Streptococcus pneumoniae is the most common bacterial cause of community acquired pneumonia (CA-PNA) in children [1]. The World Health Association (WHO) estimated in 2008 that pneumococcal disease was responsible for 476,000 global annual deaths amongst children < 5 years of age in 2008 [2]. There are more than 90 described serotypes of *Streptococcus pneumoniae* serotypes but less than 20 are responsible for the vast majority of disease. The most common serotypes were included in the initial 7-valent conjugated pneumococcal vaccine (PCV-7) which helped reduce pneumonia associated hospitalizations by 39% [3–5]. The current trend in *Streptococcus pneumoniae* infections has been the rise of multi-drug resistance in the last two decades. Moreover, there continue to be reports of outbreaks of multiple resistant pneumococcal diseases in day-care centers, shelters, and jail institutions [6–10].

In this report, we present a pair of 16 month-old African-American fraternal twins that presented with CA-PNA by penicillin-resistant *Streptococcus pneumoniae*. The pneumonia affected both twins in the same way, but a tomography scan did not reveal any shared anatomical abnormalities to account for this near-identical progression. We believe that these cases are first reported of fraternal twins with simultaneous and nearly identical illnesses due to S. pneumonia.

Case

16 month-old African-American twins presented to the ER with a 10 day history of cough. Two days prior to presentation they had been seen at their pediatrician's office. At the time, chest x-rays were normal. The pediatrician prescribed cefdinir after diagnosing the children with post-viral pneumonia on clinical grounds. Over the next two days their coughs worsened and they both became febrile and dyspneic. On the day of admission, they were again seen in their pediatrician's office where repeat x-rays were performed and they were referred to the emergency room for evaluation and treatment.

In retrospect, twin A had fever with upper respiratory symptoms and otitis media that had been treated with oral amoxicillin 2 weeks prior to admission.

Birth history revealed that the patients were full term non-identical twins born to a G1P2 mother who had appropriate prenatal care including receiving pneumococcal 13 valent vaccine. They lived at home with their mother, attending daycare. Their immunization statuses were up to date. There was no family history of asthma, but the twins had multiple food allergies including cow milk protein allergy as infants.

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Hospital course

Twin A

Upon admission, vital signs were: temperature: 103.7 F; respiratory rate: 32 bpm; oxygen saturation: 93% in room air. A physical exam showed an irritable, crying toddler. The tympanic membranes were red and landmarks were not visualized. Breath sounds were decreased bilaterally without crackles or wheezes, but the child had diffuse rhonchi with mild intercostal retraction. The remainder of the exam was unremarkable.

Laboratory investigations showed a white blood cell count of 27,400 and a C-reactive protein of 29.1 mg/L. The chest x-ray at the pediatrician's office had showed large left-sided pleural effusion with compression atelectasis of the left lung field.

The patient was placed empirically on ceftriaxone and azithromycin. Supplemental oxygen was delivered via a nasal cannula. A CT scan of the chest the following day demonstrated complete opacification of the left hemithorax. Azithromycin was stopped and vancomycin was added to his antibiotic regimen. The patient subsequently on hospital day 3 underwent video assisted thoracoscopic thorascopic decortication of the left side with pleural and visceral peel and chest tube placement.

The thick fibrinous exudate was sent for culture and yielded *Streptococcus pneumoniae* which was sensitive to vancomycin and rifampin but resistant to erythromycin, oxacillin, penicillin, and cefotaxime. The pneumococcus strain was not serotyped. Blood cultures as well as endotracheal tube cultures for acid fast bacilli and anaerobic cultures were negative.

The patient remained on vancomycin and rifampin intravenously to enhance tissue penetration. Follow-up chest x-rays showed a gradual resolution of left sided infiltrates and pleural effusion. The chest tube was removed 4 days after resolution of the pleural effusion without any complications. Quantitative immunoglobulins, total complement levels, C3, C4, and HIV tests were all normal. A purified protein derivative skin test (PPD) was also negative. The patient defervesced after 9 days and was sent home with twin B on the 12th hospital day with a PICC line for vancomycin and oral rifampin for 3 more weeks.

Twin B

Upon admission, vital signs were. temperature: 102.7 F; respiratory rate 32 bpm; oxygen saturation 93% in room air. The patient was crying and irritable. The tympanic membranes were red and landmarks were not visualized. A pulmonary evaluation demonstrated mild intercostal retractions, decreased breath sounds, diffused rhonchi, and end expiratory wheezes. The remainder of the examination was unremarkable.

Laboratory investigations showed a white blood cell count of 18,900 and a C-reactive protein of 20.2 mg/L. The chest x-ray showed a left-sided infiltrate with effusion.

A CT scan of the chest obtained the following day showed complete opacification of the left hemithorax secondary to effusion. The patient underwent the same hospital course for antimicrobials and thoracoscopic decortication with chest tube placement and removal as twin A. The fibrinous exudates also grew *Streptococcus pneumoniae* with the same antimicrobial sensitivities. Blood cultures and endotracheal tube aspirate for anaerobic and acid fast bacilli cultures were also negative. The immunologic work-up, HIV test, and PPD placement were all normal. The patient was also sent home on the same day as twin A with a PICC line and vancomycin and oral rifampin for 3 more weeks

Discussion

In a review of literature and case reports, there are no reported cases of fraternal twins with simultaneous or non-simultaneous CA-PNA or effusions. There is one case report of simultaneous occult pneumococcal bacteremia in identical twins [11] and bacteremic pneumococcal pneumonia in siblings [12]. In both cases, the conclusion was made that the infection came from a single bacterial clone and was passed down from sibling to sibling. Although it could be postulated that the infection derived from colonization obtained at daycare center, it is unclear how the twins developed the infection on the same side and progressed to the same state of pleural effusion.

There has been few reported cases of pneumonia and effusions in twins caused by other pathogens. A set of identical adult twins with fatal adenoviral pneumonia has been reported [13]. Siblings have diagnosed with Pneumococcal pneumonia [14] and pneumonia secondary to *Pneumocystis jirovecii*. [15,16]. There are two cases of siblings developing *Pneumocystis carinii* pneumonia, but the patients developed the pneumonia at different times and showed different symptoms [17]. In the immunocompromised population, a case of HIV positive infant twins who presented with *Pneumocystis* pneumonia at the same time was also reported, [18]. There also been reports a case *Bordetella* pertussis pneumonia in monozygotic twins with C3 deficiency [19]. However, unlike those in these reported cases, the patients in this report were immunocompetent, based on our routine laboratory studies, and had an almost identical presentation despite their inherent genetic differences.

One possibility based on human genetic theory of infectious diseases in which a single-gene inborn errors of immunity could be the underlying pathology in our patients. Casanova has extensively described this theory in which a single gene inborn error of immunity that has non-Mendelian inheritance and incomplete penetrance could lead to a change in the pathogenesis of infection [20,21]. For example, he described an increased in mutations of the genes encoding IL1R–associated kinase4 (IRAK4) and myeloid differentiation primary response gene 88 (MyD88) in children that has invasive pnemoccocal infections. The mutations described lead to a poor to no clinical or biological inflammatory response to pnemoccocal. Otherwise, the patients were healthy patients [21]. Unfortunately, at the time of presentation with our patient's there was no suspicion to carry out a gene-wide testing of the siblings.

It is also possible, but unlikely that the twins had identical anatomic abnormalities which led to their outcomes. However, no evidence of anatomic abnormalities was visualized on their chest computed tomography scans. The twins were non-identical twins so anatomic abnormalities are unlikely secondary to their genetic make-up.

This case of simultaneous *Streptococcus pneumoniae* in fraternal twins is remarkable in the similar course the infection took in both patients. The cause of this similar progression is still unknown as no anatomic abnormalities were detected from the patients' tomography scans. This case suggests that there may be possible similarities in fraternal twins and siblings which are not evident in routine scans that may lead to near-identical progressions of bacterial pneumonia. The cause of the simultaneous and similar infection remains unknown.

Conflict of interest

All authors have no conflict of interest to disclose.

Informed consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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