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Pediatric pneumothorax: Case studies and review of current literature

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ABSTRACT

Pneumothorax is an abnormal collection of air between the lung and chest wall. Pneumothorax management guidelines put forth by the American College of Chest Physicians, European Respiratory Society, and British Thoracic Society are specific to adult patients. These guidelines' utility has not been addressed in pediatric populations, which causes significant management variation in younger patients. Additionally, pneumothorax management ranges from conservative to surgical treatment, but these approaches, timelines, and effectiveness have not been validated in significant numbers of pediatric patients. Here, we present three cases of pediatric pneumothorax with variable clinical courses—one with persistent air leak despite chest tube presence who underwent VATS and surgical resection of apical blebs. In contrast, the other two cases were managed more conservatively. We reviewed the current literature for diagnostic and management of pediatric patients with pneumothorax, which underscores the need for guidelines specific to this population.

Educational aims

The reader will come to:

- Understand that having a unified definition of small and large pneumothorax in the pediatric population will be beneficial to further determine management plans.
- Value the need for guidelines to obtain a chest CT scan in the evaluation of spontaneous pneumothorax (SP) to evaluate for underlying lung disease such as pleural blebs
- Appreciate the importance of a Pediatric Pulmonology consult in all patients admitted to the hospital with SP, as untreated asthma can rarely be the cause of pneumothorax.

Future research directions

- Consensus should be developed regarding the management of pneumothorax in the pediatric population. This can be done by performing large multi-center prospective studies and analyzing the outcomes of different management approaches. However, a standardized definition of the size of the pneumothorax should be attained.

1. Introduction

Pneumothorax is an abnormal collection of air in the pleural space between the lung and chest wall. Pneumothorax is classified as either traumatic or spontaneous. Traumatic pneumothorax is a consequence of blunt or sharp trauma to the chest, mechanical ventilation, and diagnostic or therapeutic procedures. Spontaneous pneumothorax (SP), which occurs without an obvious etiology, is classified as primary spontaneous pneumothorax (PSP) and secondary spontaneous pneumothorax (SSP). PSP occurs in patients without known underlying disease, while SSP occurs in patients with conditions that affect the lung parenchyma, such as infectious pneumonia, interstitial lung diseases, Marfan syndrome, and connective tissue disorders, or disorders of the airways such as asthma [1]. In the pediatric population, other factors have also been implicated, for example, cystic fibrosis, necrotizing pneumonia, pneumatoceles of any cause, congenital lobar emphysema, and immunodeficiency [2]. A recent 12-year retrospective cohort study found that children with asthma have a greater risk of pneumothorax than children without asthma. The hazard ratio of pneumothorax in

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Abbreviations

ACCP	American College of Chest Physicians
BTS	British thoracic society
CXR	chest x-ray
EHR	electronic health record
ERS	European Respiratory Society
SP	spontaneous pneumothorax
PSP	primary spontaneous pneumothorax
SSP	secondary spontaneous pneumothorax
VATS	Video-assisted thoracoscopic surgery
IRB	Institutional review board
PFT	pulmonary function test
PJP	Pneumocystis Jerovecii

asthmatic children was higher among those with poorly controlled asthma [3]. Pediatric SP has been noticed to occur at rest rather than with exertion and can be precipitated by straining, lifting, or other Valsalva maneuvers [4]. Male sex, tall and thin body habitus, adolescence, and young adulthood have all been described as risk factors for pneumothorax [2,5]. Another entity called catamenial pneumothorax has been described in the literature, which is a recurrent pneumothorax in women of reproductive age and has been associated with the thoracic manifestation of endometriosis [6].

PSP has an estimated incidence of 7.4–18 cases per 100,000 population per year among men and about 1.2–6 cases per 100,000 population per year among women [7]. The incidence of SSP is similar to that of PSP, also more predominant in males [7]. In the pediatric population, pneumothorax has been reported to occur in 4 per 100,000 males and 1.1 per 100,000 females per year, suggesting a lower incidence in children than adults [7]. However, it has been reported to have a higher recurrence rate in the pediatric population [8,9].

The most common symptoms of SP are acute onset of chest pain with shortness of breath, with respiratory distress being less common. Symptoms are also accompanied by pleuritic chest pain, which can evolve into dull achy pain in a couple of days [7]. Commonly seen physical exam findings are diminished breath sounds, dyspnea, tachycardia, hyper-resonant percussion, and decreased vocal fremitus. The severity of presentation is also dependent on the size of the pneumothorax.

American College of Chest Physicians (ACCP), European Respiratory Society (ERS), and British Thoracic Society (BTS) have published guidelines for the management of pneumothorax in the adult population. Still, there are no clear guidelines for managing pneumothorax in the pediatric population, and most of the management decisions are extrapolated from the adult guidelines [7].

We present three patients who were hospitalized with spontaneous pneumothorax and had different clinical courses and managements. We comment on the possible causes of pneumothorax as this could affect the recurrence. We reviewed the literature and current guidelines for diagnosis and management specific to the pediatric population.

2. Methods

We reviewed clinical presentation, imaging studies, and management of three patients admitted to our pediatric service with a diagnosis of spontaneous pneumothorax between June–December 2020. Institutional review board (IRB) approval was not needed for this case series.

2.1. Case 1

A 16-year-old male with a history of intermittent asthma presented to the ED with sudden onset of dyspnea and chest pain. His symptoms had progressed rapidly over 1 hour to severe dyspnea, for which his mother had called EMS. There was no associated fever, rhinorrhea, cough, vomiting, or retching. He tried anti-reflux medication and Albuterol at home, with no response. EMS gave him a dose of solumedrol en route to the ED. In ED, he received NSAIDs for pain, and his chest x-ray (CXR) revealed a left pneumothorax with a mild rightward shift of the mediastinum (Fig. 1A). A chest tube was placed in the ED with full re-expansion of the left lung. He was admitted to the PICU, where he remained stable, with a left chest tube placed to continuous suction at −20 cmH2O and adequate pain control. On day two of admission, CXR showed re-expansion of the left lung without abnormalities in the lung parenchyma. His physical exam was within normal limits, with symmetric chest expansion and clear breath sounds bilaterally.

Further review of his past medical history revealed a history of mild intermittent asthma, for which he used to take Albuterol as needed. He had no prior history of hospital admissions for asthma or any other respiratory illness. Pediatric Pulmonology was consulted and made recommendations to start him on maintenance inhaled corticosteroids for controlling his asthma. They also suggested PFT in an outpatient setting for evaluation of his asthma.

On day three of hospital admission, the chest tube was changed to water seal, but after a few hours, he started complaining of an increase in chest pain, and a CXR revealed re-accumulation of air on the left side. The chest tube was switched back to continuous suction at −20 cmH2O, and a repeat CXR showed the resolution of left pneumothorax.

On day 4 of admission, continued air bubbles were noticed from the chest tube while still at suction of −20 cm H2O. As the air leak persisted, a chest CT scan was performed, which showed bilateral apical blebs and the pneumothorax on the left side (Fig. 1B and C).

The surgical team was consulted. He underwent left-sided video-assisted thoracoscopic surgery (VATS) with left apical pulmonary wedge resection and chemical and mechanical pleurodesis on day five of admission (Fig. 1D). Although bilateral blebs were seen on chest CT, it was recommended to withhold intervention on the right lung. For the next three days, the patient continued to have air bubbles through the left chest tube. The Chest tube was placed on water seal on day 9 of admission when there was no more air leak and was removed the next day. However, a few hours later, the patient started complaining of chest pain, difficulty breathing. A CXR showed a recurrent pneumothorax on the left side. He was taken to radiology for emergent interventional radiology (IR) guided chest tube insertion overnight. He was monitored for the next few days until no further leak was noticed. The chest tube was successfully removed on day 14 of admission, and he was discharged home on day 15 with a normal CXR. Recommendations were made for close follow-up with pediatric pulmonology and surgery. During follow-up visits, CXRs have been normal except for postoperative changes. The family was provided the option of right-sided elective apical wedge resection to prevent future recurrence of pneumothorax.

2.2. Case 2

A 9-year-old female with a history of mild intermittent asthma, and no prior hospitalization, presented to ED with a two-day history of headache and one-day history of shortness of breath that did not respond to albuterol administration at home. In the ED, she was found to have a right-sided moderate pneumothorax and pneumomediastinum on CXR (Fig. 2A). Her nasal swab was positive for Influenza A, and she was started on oseltamivir.

She was admitted to the PICU and remained hemodynamically stable but with mild respiratory distress accompanied by biphasic wheezing and poor aeration over the right lung. She underwent chest tube placement and repeat CXR demonstrated interval improvement of the pneumothorax. The chest tube was initially placed to −20 cm H2O suction. Pediatric Pulmonology was consulted, who recommended bronchodilators and systemic steroids for the management of acute asthma exacerbation. Her bronchodilators and systemic steroids were

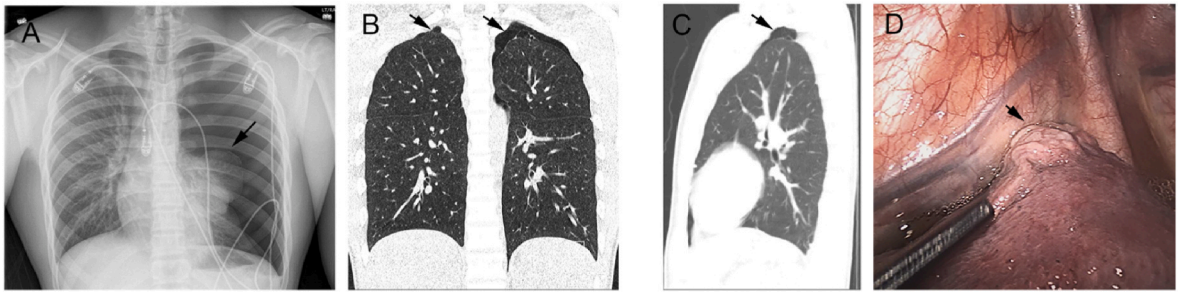

Fig. 1. (**A**) CXR on day 1 shows left pneumothorax and lung collapse (arrow), with mild rightward shift of the mediastinum. (**B, C**) Chest CT scan on day 4 showing bilateral apical blebs and left pneumothorax (arrow), (**D**) Apical blebs visualized during VATS (arrow).

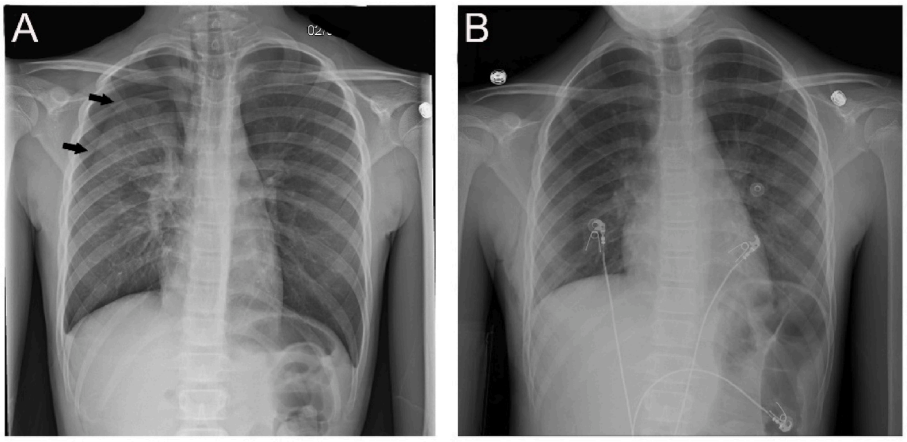

Fig. 2. (**A**) CXR on day 1 showing right pneumothorax (arrow) and pneumomediastinum. (**B**) CXR on day 4 showing resolution of pneumothorax.

continued for five days and progressively weaned off. The chest tube was removed on day 4 when no air bubbles were observed under the water seal, and CXR showed lung re-expansion (Fig. 2B).

On further evaluation by Pediatric Pulmonology, the family reported that the patient had been coughing at baseline, suggestive of persistent asthma. Spirometry was obtained 2 weeks after discharge and showed a reversible obstructive pattern. She was started on daily inhaled corticosteroids as a controller medication for asthma. During her follow-up, a chest CT scan was performed to evaluate for any underlying lung pathology, which was normal.

2.3. Case 3

A 15-year-old male presented to the ED with one day history of dyspnea and chest pain. A CXR revealed a left-sided pneumothorax (Fig. 3A and B). He was started on oxygen via a non-rebreather mask in the ED. The pediatric surgery team was consulted, and he was admitted to the hospital. He was continued on oxygen via a non-rebreather mask and monitored for changes in respiratory status. After approximately 24 hours, his dyspnea and chest pain worsened, and the size of pneumothorax increased on CXR. On day 2 of hospital admission, a chest tube was inserted on the left side. His symptoms improved, and a CXR revealed good expansion of the left lung. The chest tube remained in place for three days, then removed after observing no air bubbles under the water seal. Repeat CXR did not show any pneumothorax after chest tube removal on day 4 of admission when he was discharged home. (Fig. 3C). Further chart review revealed that he had been previously prescribed Albuterol for intermittent wheezing.

3. Discussion

Pneumothorax can present with different degrees of severity and is a

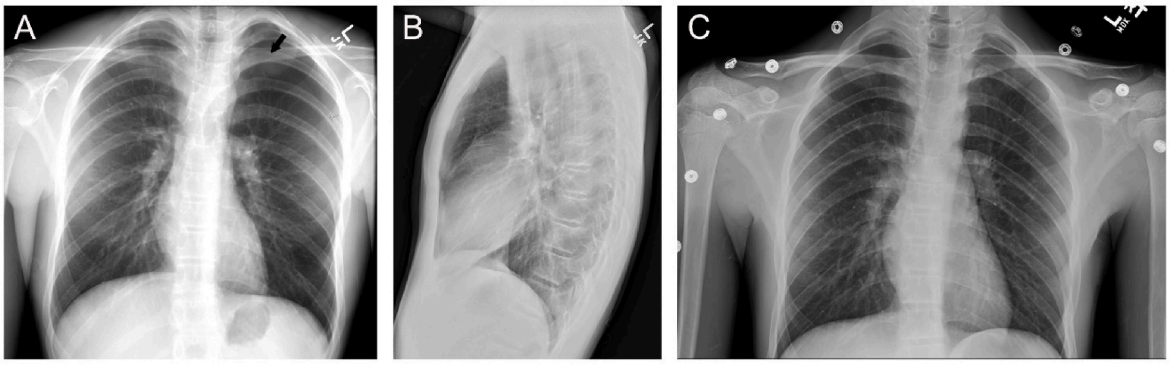

Fig. 3. (**A, B**) CXR with PA and lateral views on day1 showing left pneumothorax (arrow). (**C**) CXR on day 4 showing resolution of pneumothorax.

potentially life-threatening condition. Management guidelines for pneumothorax by the ACCP, ERS, and BTS are used as a reference in the adult population. It is imperative to emphasize that none of these guidelines discuss specific applications in the pediatric population [7].

In this case series, we aimed to emphasize the importance of investigating the underlying etiology of pneumothorax in pediatric patients. As discussed above, the patient in case 1 had a history of intermittent asthma, which reportedly was well controlled but was found to have apical blebs as an underlying cause of his pneumothorax. CT scan and surgical consultation were prompted due to a persistent air leak despite having a chest tube on the affected side for 4 days. In contrast, the patient in case 2 was treated with systemic steroids and bronchodilators for acute asthma exacerbation during her hospitalization. During her pulmonology follow-up, she continued to present clinical and spirometry evidence of poorly controlled asthma and was started on controller therapy with inhaled corticosteroids. The patient in case 3 had a history of occasional albuterol use. However, while he was hospitalized, the Pediatric Pulmonology service was not consulted. No investigations for underlying lung disease as a possible cause of spontaneous pneumothorax were performed. Blebs may be present for a long time before they rupture. The presence of blebs or bullae on a CT scan during the first episode of PSP can predispose to future ipsilateral or contralateral pneumothorax [10]. A recent study documented a high incidence of bullae and blebs in post mortem CT scans of healthy adults without any prior history of pulmonary disease [11]. Therefore, it is important to stress on the pulmonology follow-up because it can help decrease the recurrence by offering preventive management approaches [12].

Pneumothorax is considered a potential complication of obstructive lung diseases, including COPD and severe asthma [1,13]. Several case series and case reports have indicated difficulties with asthma management and increased recurrence of asthma exacerbations due to underlying pneumothorax [14]. Some studies have also recommended that getting a chest x-ray in a hospitalized patient with severe asthma can help detect underlying pneumothorax or pneumomediastinum. Hyperinflation and increased intrathoracic pressure can predispose to pneumothorax development. Therefore, it is important to address asthma with bronchodilators and systemic steroids in addition to pneumothorax management [14]. Pneumothorax is a rare complication of asthma and has been traditionally classified under SSP or Tension pneumothorax depending upon the clinical picture. In most patients with asthma, the pneumothorax is small and can be managed conservatively. Previous guidelines have suggested chest tube placement for management of SSP or tension pneumothorax as compared to observation in asymptomatic PSP due to complexities of underlying lung disease [2]. A recent study described that children with poorly controlled asthma are more prone to have pneumothorax as compared to those with well-controlled asthma [15].

Pneumothorax associated with cystic fibrosis has been previously associated with poor transplant outcomes and requires early surgical management. Management of HIV related pneumothorax is more focused towards the use of antiviral and prevention of Pneumocystis Jerovecii (PJP) by appropriate prophylaxis. Surgical approaches, including plication of the diaphragm, mesh repair, and electrocoagulation of endometriosis for management of catamenial pneumothorax, have not been very fruitful to prevent recurrence unless used in combination with gonadotropin-releasing hormone analogues [2].

For the management of pneumothorax, a stepwise approach is used depending upon the presentation and size of the pneumothorax. The most used strategies are presented in Table 1. Management guidelines differ in terms of classifying the degree of lung collapse and opinions regarding the use of simple needle aspiration in PSP [16]. ACCP guidelines consider a pneumothorax to be large when it extends to more than 3 cm from the chest wall apex to the ipsilateral apex of the lung parenchyma on an upright CXR. ERS suggests that management does not solely depend on the pneumothorax size but also verifies that there is no accurate method to measure it precisely. In contrast, according to BTS guidelines, a pneumothorax is considered large when it is greater than or equal to 2 cm [2,16,17]. A solid classification of pediatric pneumothorax is needed due to variations of the chest size and variation in treatment modalities which highly depend on pneumothorax size [7].

CXR has been suggested as a preferred method for diagnosis of pneumothorax by BTS, ERS, and ACCP, but the utility of chest CT scan cannot be ignored. Chest CT scan is better than CXR in calculating the size of pneumothorax and should be used as a preferred imaging modality in complicated pneumothorax, recurrent or persistent pneumothorax, and those patients being evaluated for surgical management [2,16,17].

The use of lung ultrasound for the detection of pneumothorax in adult trauma patients has shown increased sensitivity as compared to CXR, with a negative predictive value of 93.4% [18]. It has been promoted as an essential tool for pneumothorax detection in the pediatric ER without causing significant radiation exposure [19]. The absence of lung sliding is an important sign in the detection of pneumothorax on ultrasound. Still, its utility has also been questioned specifically in conditions where lung sliding sign is absent due to underlying lung diseases, including phrenic nerve paralysis, pulmonary contusion, pulmonary adhesions, and blebs [20].

Persistent air leak has been defined differently in previous studies ranging from 48 hours to more than 10 days [21], but an arbitrary cut-off point of 5 days has been suggested as well [21]. The size and number of bullae in SP have been important risk factors for recurrence [22]. The recurrence risk increases by 75% if these bullae or blebs are present bilaterally, but the CT scan's timing and utility for diagnosing them are still debatable [10]. CT scan can be utilized to detect small pneumothorax with or without bullous lung disease [2]. Performing an early CT scan has been helpful in detecting underlying lung disease, including apical emphysematous lung changes in about 60% of PSP [23]. Since most blebs are confined to the lung apex, a modified CT scan has also been suggested to help detect them with minimal radiation exposure [24].

For PSP management, ACCP guidelines suggest observation in the Emergency Department for clinically stable patients with small

Table 1
Management options for pediatric pneumothorax.

Intervention	Indication	Outcome	Reference
Observation	Used in clinically stable patients with small pneumothorax RCT of patients with moderate-to-large primary pneumothorax	It took as long as 32 days for pneumothorax to resolve in pediatric patients Observation was not inferior to interventional management	[4,28]
Oxygen	Supplemental oxygen used at high partial pressures to improve air reabsorption from pleural cavity in asymptomatic patients with small pneumothorax	4 times increase in reabsorption rate if pneumothorax>30% and a 2-fold increase in reabsorption if < 30%	[4,26]
Needle aspiration	Symptomatic small pneumothorax	Has been helpful in pneumothorax <30%, but the recurrence rate is higher	[21]
Chest tube insertion	first line for symptomatic patients or patients with large pneumothorax	The recurrence rate is higher as compared to surgical intervention	[26]
Surgical management	VATS or thoracotomies with chemical pleurodesis for persistent air leaks	Not used as the first-line but recurrence rate was less than chest tube insertion and needle aspiration	[2,16]

pneumothorax. Whereas BTS guidelines additionally recommend observing asymptomatic patients even with large pneumothorax [2,4]. ERS also supports the observation of asymptomatic patients regardless of the size [17]. Although the failure of needle aspiration in PSP ranges between 25% and 50%, BTS and ERS recommend needle aspiration as the first step in managing symptomatic PSP, whereas ACCP recommends chest tube placement in those [2,16,17]. Both ACCP and BTS guidelines suggest surgical evaluation for persistent air leak or recurrent pneumothorax in patients who already have a chest tube for 3–5 days [2,4]. For SSP management, adult guidelines recommend placing a chest tube in the presence of large pneumothorax, which can be extrapolated to the pediatric population [7].

Needle aspiration is a less invasive method to treat pneumothorax as compared to chest tube placement and has been successfully used at some centers for managing pneumothorax <30% in size in pediatric and adult patients [21,25]. A recent meta-analysis of pediatric pneumothorax management stated that PSP patients managed with non-surgical strategies such as needle aspiration and chest tube have a higher risk of recurrence compared to surgical intervention like thoracotomy, thoracoscopic surgery with blebectomy, or apicectomy with pleurodesis [26]. Increased recurrence rate (37%–52%) was noticed in patients with SP in the presence of blebs/bullae, suggesting it is a significant risk factor for recurrence [27]. Surgical options, including VATS with pleurodesis and thoracotomy, have been recommended in the management of persistent air leaks in SSP or recurrent PSP and have been successful in preventing recurrence as compared to non-surgical management [2,4,17]. A large, prospective, open-label, multicenter, noninferiority trial recruited patients with uncomplicated, unilateral, moderate-to-large primary spontaneous pneumothorax and randomly assigned them to either observation or interventional management with chest tube placement group. The investigators concluded that the lung reexpansion rate was only slightly lower than the intervention group. At the same time, conservative management resulted in shorter hospital stay and fewer adverse events [28].

Using large-bore chest tubes versus small-bore pigtail catheters as an initial treatment of choice is also controversial. ACCP recommends using a chest tube (16–26 fr) for large and unstable primary and secondary pneumothorax. However, BTS and ERS favor the use of small-bore chest drain [2,16,17]. A recent systematic review compared 11 studies and found that the duration of hospital stays and chest tube drainage is shorter in small-bore pigtail catheters versus large-bore chest tubes. Other confounding factors that could impact the duration of hospital stay and chest tube removal were not discussed at length in that review [29]. Overall, the findings of this systematic review are in congruence with a pediatric study, which concluded that a small-bore pigtail catheter is a safe alternative to large-bore chest tubes in neonates with the additional advantage of requiring less time to perform the procedure [30].

VATS with apical blebectomy and pleurodesis has been a preferred approach for both recurrent PSP or persistent air leaks with initial PSP [31.] In children, SP can be caused by the rupture of bullae or blebs, which can be acquired or congenital [21]. Early surgical treatment, within 48 hours, has been suggested to improve the outcomes of recurrent pneumothorax in the pediatric population compared to adults [21]. These findings suggest that obtaining a chest CT scan during the admission can be helpful not only for diagnosis of underlying lung or airway pathology that can lead to pneumothorax but also identifying other anomalies such as bronchopulmonary fistula, congenital pulmonary and airway malformations, which can be mistaken for PSP [23].

4. Conclusion

Pneumothorax management in pediatric patients is variable due to the lack of guidelines. The management also depends on the underlying disease process. It is essential to identify the underlying disease process at the earliest presentation to improve the outcomes and decrease the recurrence of pneumothorax. Different imaging modalities, including CXR, ultrasound, and CT scan, can help diagnose underlying lung disease.

Conflict of interest/funding

None.

Declaration of competing interest

- We have no conflicts of interest, real or perceived, to disclose.
- All the authors approve the current submission.
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