Clinical Case Report

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Approach to diagnosis of factitious disorder with unexplained hemoptysis

A case report

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Abstract

Rationale: Hemoptysis or hematemesis is a common clinical symptom in adults, but is unusually to be observed in children. Hemoptysis can occur with circulatory diseases, injuries, several types of systemic diseases, or systemic factors including factitious disorder (FD), which is difficult to be diagnosed. Here, we report a case of hemoptysis caused by FD to provide a diagnostic flow chart for such kind of disease.

Patient concerns: An 11-year-old female patient had a history of hemoptysis or hematemesis for 6 months and suffered with paroxysmal syncope for a month.

Diagnosis and intervention: A series of examinations had been launched to evaluate any possible malformation or abnormalities of the patient including fiberoptic bronchoscopy, cardiac catheterization, gastroscopy, nasolaryngoscopy, electrocardiogram, electroencephalogram, and enhanced magnetic resonance image of the paranasal sinus. Several methods had been performed and tried to stop hemoptysis such as taking hemostatic medications, lavage of fiberoptic bronchoscopy, and embolism for abnormal bronchial arterial using cardiac catheter. All the interventions, however, failed to achieve our treatment goal. Given that more careful observation during hospitalization had been done, and we suspected the symptom of hemoptysis from this patient might be originated from an FD.

Outcomes: Based on the diagnosis of FD, targeted psychological intervention was provided by experts. After the treatment completed, the patient did not present hemoptysis anymore.

Lessons: FD is an uncommon type of disease. This rare case described here is to help us to reconsider the long diagnosis process of hemoptysis with a series of examinations including some invasive procedures, whether all the examinations and interventions are necessary for a nonsevere hemoptysis patient.

Abbreviations: CT = computed tomography, FD = factitious disorder, MRI = magnetic resonance imaging.

Keywords: case report, child, factitious disorder, hemoptysis

1. Introduction

Hemoptysis is defined as bleeding from the respiratory organs below the larynx with the discharge of blood from the mouth through a cough. The causes of hemoptysis vary; the most common reasons are infections of the respiratory system, circulation system diseases, injuries, and systemic hemorrhagic tendency diseases. In addition, some unusual diseases can lead to hemoptysis, such as alternative menstruation, oxygen poisoning, pulmonary hemorrhage nephritis syndrome, bronchiectasis, sinusitis, and visceral translocation syndrome. Factitious disorder (FD) is an uncommon cause of hemoptysis, presenting a pattern of falsification of physical or psychological signs or symptoms, associated with identified deception.[1] Furthermore, FD is a rare disease that has only been presented in several case reports since the 1970s.[2–5] For such a population, physicians usually cannot reach a clear diagnosis in a short period of time. Patients with FD often undergo a series of interventions to treat nonmassive bleeding. However, if the patients need to be distinguished from those with Munchausen syndrome, this usually requires a longer procedure.[6,7] With the rapid development of vessel scanning and transcatheter interventions, the incidence of FD cases receiving unnecessary medical treatment. According to previous reports, each author focused on different presentations, and no one paid attention to how to clarify the diagnostic process. Here, we report a rare case of hemoptysis caused by FD to learn about it; the patient with his guardian signed an informed consent statement to allow this case to be published. Most important, this patient underwent several interventions including invasive vessel closure; therefore, we summarized the diagnostic procedures of his unexplained hemoptysis.

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Table 1

Auxiliary examination and its significance.

Inspection item	Result	Positive	Negative
Chest radiographs	No apparent abnormality		Respiratory tract infections
CT	No apparent abnormality		Bronchiectasis, chronic obstructive pulmonary disease, and malignancy
Blood coagulate functions	(2016–10–12) TT: 28s (↑) Antithrombin III: 138.8% (↑); (2016–12–08) APTT: 41.10s (↑) Contractinogen: 1.61g/l (↓) (2017–05–30) No apparent abnormality		Blood system diseases
Fiberoptic bronchoscopy	Mild inflammation of bronchial mucosa		Hemosiderin in the lungs
Echocardiography	Non apparent abnormality		Cardiovascular diseases
Cardiac catheterization	A small volume of pulmonary collateral branches	Support for hemoptysis. However, after bronchial artery embolization, the patient's condition did not improve	
Nasolaryngoscopy	No active bleeding in acute rhinitis		Nasopharyngeal hemorrhage
Gastroscopy	Chronic nonatrophic gastritis		Hemorrhage of digestive tract
MRI for paranasal sinus	Maxillary sinus cyst		Accessory sinus hemorrhage

CT = computed tomography; MRI = magnetic resonance imaging.

2. Case presentation

The child presented with recurrent hemoptysis and recent paroxysmal syncope. The patient frequently produced bright red or dark red blood substances, calculating approximately 5 to 90 mL each time, more than once a day. Nevertheless, the child did not have anemia, and denied a history of blood transfusions. Many medical examinations were carried out to identify possible causes. Chest radiograph indicated that there was no obvious pulmonary and bronchial infection. Fiberoptic bronchoscopy showed only slight inflammation in the upper lobe of the lung, and there was no hemosiderosis in the lungs. Cardiac catheterization demonstrated that there was a small vessel of pulmonary collateral branches in the right intercostal area, passing through the right 6/7 intercostal area. Multiple gastroscopies prompted chronic nonatrophic gastritis with erosion. Nasopharyngeal laryngoscopy revealed no clear abnormalities. Electrocardiogram showed sinus rhythm, electric axis unbiased, and first-degree atrioventricular block. Electroencephalogram showed a normal school-age waking pattern. There was no abnormal examination of the reproductive system by B-ultrasound. Enhanced computed tomography (CT) and magnetic resonance imaging (MRI) of paranasal sinuses were more likely to reveal submucosal cysts in bilateral maxillary sinuses; bilateral ethmoid sinus, sphenoid sinus, and frontal sinus as a normal condition; and only bilateral inferior turbinate revealed slight hypertrophy, providing no clues for hemoptysis. All examinations are listed in Table 1. Ethamsylate and hemocoagulase were used to prevent bleeding, and Losec and Colphos were used to suppress gastric acid and protect the stomach. Nevertheless, hemoptysis persisted. Syncope occurred several times a day, manifesting as binocular closure, non-responsiveness, and limpness persisting 1 hour at the longest. Body position was not related to syncope. Syncope always followed optometry and nausea. These attacks of syncope were observed to be related to mood during hospitalization. After fainting, external stimulation could help the child to wake up. It remained unclear whether syncope was a true symptom or a "drama" play. Because all measurements and treatments failed to stop the bleeding, we began to consider whether the paroxysmal symptoms were psychogenic. Furthermore, during hospitalization, the patient spat bloody fluid from the mouth. The times of episodes and the amount of blood spitting varied every day. Most of it happened at night when she was alone with her parents. During the day, the patient always went out to play with other children and reported no hemoptysis. A psychoanalyst was invited to evaluate the patient and could not provide a diagnosis of a psychological problem. Upon inquiry, there was no history of mental illness in her family. Finally, according to the diagnostic procedure of FD, the patient was believed to suffer from FD (Supplemental Table I, http://links.lww.com/MD/C783). Therefore, after a week of in-hospital observation, we advised the patient to return home and continue out-patient follow-up; we allowed her parents to pay more attention to the psychological status of the child and to employ behavior and psychological intervention on a schedule. We found that the patient's symptoms disappeared after half a year.

3. Discussion

Hemoptysis in childhood is an uncommon clinical symptom. Because of the special period of childhood, it is very difficult to obtain medical history and have the cooperation of the child for the examination, making it more difficult to identify the real cause of hemoptysis. In general, the most common cause of hemoptysis in childhood is infection, including pneumonia, tuberculosis, lung abscess, and infectious bronchiectasis. Congenital heart disease is the second largest cause of hemoptysis in children. Inhaled foreign substances, cystic fibrosis, tumor of respiratory system, idiopathic pulmonary hemosiderosis, upper airway stenosis, and causes of pulmonary hemorrhage such as systemic lupus erythematosus, Goodpasture syndrome, pulmonary thromboembolism, hydatid cyst, and even gastric duplication cyst can induce hemoptysis.[8] Solitary pulmonary arteritis can also lead to massive hemoptysis in children.[9] Amenorrhea hemoptysis is caused by endobronchial heterosis.[10] Finally, there is factitious hemoptysis.[3] Even though the causes of hemoptysis are diverse, most of the etiological factors can be distinguished by precise examinations such as CT and MRI.

The patient underwent a series of examinations and treatments (Table 1), but the true reasons for the disease remain missing. For hemoptysis patients, respiratory diseases should be considered as

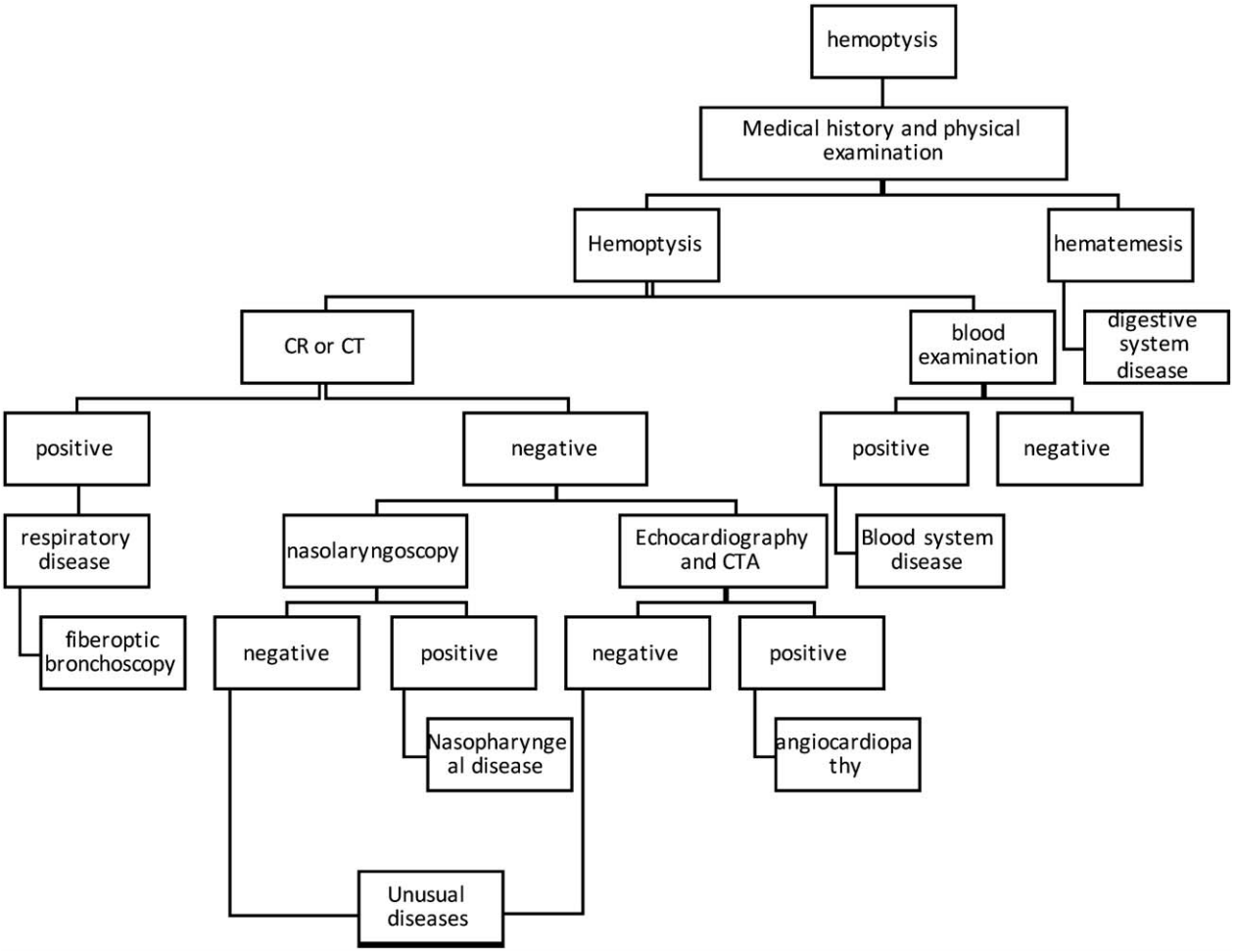

Figure 1. Flow chart for diagnosis process of unexplained hemoptysis. CR = chest radiography, CT = computed tomography.

first; however, most of them can be ruled out after chest radiographs and CT test. We performed fiberoptic bronchoscopy to exclude diseases such as hemosiderin in the lungs. Then, cardiovascular disease was evaluated by echocardiography and cardiac catheter angiography, revealing a small vessel of pulmonary collateral branches within the right intercostal area. However, after bronchial artery embolization with a device, the patient continued to suffer hemoptysis. Subsequently, we began to question whether the bleeding came from the digestive system. After nasolaryngoscopy and gastroscopy, no positive clues were identified. Furthermore, we examined the paranasal sinus with MRI and found nothing abnormal.

According to Dimsdale et al's report, FD is a long-term and persistent problem related to disease perception and identity. It may be associated with inexplicable and/or unexpected symptoms. In general, a diagnosis can be considered by meeting the following 4 criteria. First, a pattern of presentation to others as ill or impaired; second, the behavior is evident even in the absence of obvious external rewards; third, the behavior is not due to a delusional belief system or acute psychosis; and fourth, the behavior is not better accounted for by another mental disorder[11] (Supplemental Table I, http://links.lww.com/MD/C783). The case described in this report presented continuous hemoptysis. There was no obvious reward mechanism for hemoptysis during hospitalization. Furthermore, the results of our examinations did not support the patient's hemoptysis. The point is that his mental state was not obviously abnormal (Supplemental Table I, http://links.lww.com/MD/C783). Therefore, we considered that the diagnosis of FD was acceptable. According to the results of our follow-up, our psychological intervention was effective, meaning our diagnosis was reliable.

However, for this patient, during hospital stay, a series of examinations was performed, including some invasive ones, and catheter interventional treatment was performed for suspected abnormal vessels. This caused us a re-evaluate whether we performed too many examinations for a nonsevere symptom in a child. Therefore, based on this case, we created a flow chart for unusual hemoptysis diagnoses to avoid excessive examinations (Fig. 1).

This case report still has several limitations. First, we did not find any tools or methods that the child used to harm himself. Therefore, the diagnosis was reached mainly based on the outcomes from attempts at behavioral and psychological intervention. Second, this is only a case report; therefore, the suggested diagnostic procedure needs to be validated in a case series.

Author contributions

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Writing – review and editing: Yimin Hua, Jie Fang, Yifei Li.

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