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PEDIATRIC RADIOLOGY

ORIGINAL ARTICLE

Consolidation in the right middle lobe in pediatric bronchial–pulmonary artery shunt: radiology's Aunt Minnie?

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PURPOSE

By retrospectively studying the chest computed tomography (CT) data of children with bronchial artery (BA)–pulmonary artery fistula, this study summarizes the characteristic imaging features of the disease and provides imaging support for the diagnosis and clinical treatment of these children.

METHODS

Digital subtraction angiography and CT angiography data were collected from 74 children with pulmonary hemorrhage following BA embolization. Bronchial–pulmonary shunt was present in 30 cases.

RESULTS

Of the 74 children with pulmonary hemorrhage in this study, seven exhibited signs of consolidation in the middle lobe of the right lung, and bronchial–pulmonary shunt existed in all of them. A total of 30 children with BA–pulmonary artery shunt (PAS) had BA tortuosity and thickening. Regarding primary BA-PAS, the middle lobe and lower lobe of the right lung were involved in 94.1% (16) of the children. Those with a fistula located in the middle lobe of the right lung accounted for 58.8% (10 cases), of which 40.0% (four cases) presented consolidation. In this study, 41.2% (seven) of the children with primary BA-PAS exhibited no abnormal changes on chest CT, and 58.8% (10 cases) exhibited abnormal changes in the unilateral lung.

CONCLUSION

For children with pulmonary hemorrhage who have consolidation in the right middle lobe, the formation of BA-PAS should be anticipated. The possibility of primary BA-PAS should not be disregarded in children with pulmonary hemorrhage with tortuosity and dilation of BAs, despite no apparent abnormalities on lung CT, or ground-glass density or consolidation on only one side.

CLINICAL SIGNIFICANCE

The chest CT of patients with pulmonary hemorrhage showed consolidation of the right middle lobe of the lung, which was highly likely to indicate BA-PAS.

KEYWORDS

Computed tomography, X-ray, child, bronchial artery–pulmonary artery shunt

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Pulmonary hemorrhage occurs when blood enters the alveoli or airways, which is rare and may be insidious among children.[1] Clinical symptoms in children are variable and can range from asymptomatic to bloody sputum or hemoptysis.[2] However, it is challenging to detect pulmonary hemorrhage or hemoptysis in younger children because they usually swallow bloody sputum or because the bleeding is minimal. Common causes of pulmonary hemorrhage in children include idiopathic hemosiderosis, pulmonary cystic fibrosis, pulmonary vascular malformation, primary heart disease, hematological disease, and tumors.[3]

The bronchial artery (BA) arises from the T5–6 level of the thoracic aorta. The BAs outside the descending thoracic aorta at the T5–6 level are defined as ectopic BAs, also known as vagal

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BAs.[4] BAs normally receive only approximately 1% of the heart's blood output to maintain airway and lung function. They supply blood to the bronchi, esophagus, visceral pleura, thoracic aorta, pulmonary arteries, nerves, pulmonary veins, and lymph nodes in the thoracic cavity.[5] They do not normally participate in gas exchange. The BA is connected to the pulmonary artery by anastomosis of several microvessels at the level of the alveolar and respiratory bronchioles. In children with pulmonary hemorrhage, the BA and its anastomotic site may dilate to form a BA–pulmonary artery shunt (PAS),[6] also termed bronchial–pulmonary arterial fistula,[7] which further exacerbates bleeding symptoms. The normal pressure in the pulmonary artery is approximately 13 mmHg, whereas the pressure in the BA can be as high as 95 mmHg. The marked difference in pressure leads to the rupture of the BA-PAS, causing pulmonary hemorrhage. A BA-PAS can also be primary, and some researchers believe that it is related to fetal hypoxia in utero.[8] Primary BA-PAS is an exclusive diagnosis in the clinic. Children with primary BA-PAS are often untreated and require BA embolization to improve clinical symptoms. Bronchial arteriography is an invasive test that is often performed in patients with massive hemoptysis.[9] The interventional treatment for primary and secondary BA-PAS is the same. The formation of BA-PAS is a major indication of BA embolization.[10]

Early detection of BA-PAS on computed tomography (CT) and timely BA embolization can effectively alleviate patients' symptoms and improve their prognosis. We hypothesize that there are characteristic findings on CT imaging that increase the probability of BA-PAS diagnosis in children that may otherwise be missed. We thus reviewed 74 BA-PAS cases from the National Medical Center of Pediatrics in China. If our hypothesis is proven to be correct, children with BA-PAS could benefit from this study.

Main points

- Chest computed tomography (CT) images of pulmonary hemorrhage in children are varied and may even be negative.
- Primary bronchial artery–pulmonary artery shunt (BA-PAS) typically manifests as ground-glass opacity or consolidation in one side of the lung.
- Not all patients with BA-PAS develop right middle lobe consolidation.
- The formation of BA-PAS should be considered when the right middle lobe of the lung is seen on chest CT in children with pulmonary hemorrhage.

Methods

Data acquisition

This study was a single-center retrospective observational study conducted between March 1, 2016 and March 31, 2020. The study was approved by the Ethics Committee of Beijing Children's Hospital (decision no: IEC-C-008-A08-V.05.1, date: 25.12.2018). Being a retrospective study, it was approved by the ethics committee without informed consent.

The CT images and clinical data of 334 children with pulmonary hemorrhage were reviewed. A total of 74 children who underwent bronchial arteriography for pulmonary hemorrhage were enrolled in the study. The inclusion criteria were as follows: (1) clinical diagnosis of pulmonary hemorrhage; (2) patients who underwent chest CT; (3) patients who underwent BA digital subtraction angiography (DSA); and (4) patients who underwent chest CT angiography (CTA). The exclusion criteria included: (1) patients with pulmonary hemorrhage of unclear etiology; (2) poor-quality CT scan images for evaluation; (3) patients with poor adherence or unable to cooperate adequately with the examination because of their illness or other reasons; and (4) patients with lung contusion and laceration.

Scan protocol

All DSA scans were performed using a Siemens AXIOM Artis. Patients were under general anesthesia during the operation, where a percutaneous femoral artery puncture using a modified Seldinger technique was performed. Super-selective contrast was administered using the hand method (10–15 mL, 2–3 mL/s). The contrast agent was non-ionic (Ultravist® 370). Once the location of the BA was established, super-selective intubation was performed using a 3F coaxial microcatheter on a guidewire first principle. Angiography was performed repeatedly, and important branches, such as the spinal and esophageal arteries, were avoided during the procedure. The main size of the embolization material was approximately 300–500 μm, and the size of the selected particles was adjusted appropriately according to whether there was an arteriovenous fistula. If the BA shares a trunk with the internal thoracic artery, protective embolization of the distal artery can be performed using a spring coil. Following the manual identification of the BA, angiography was performed using a high-pressure syringe. The presence of a pulmonary artery image in the arterial phase indicated BA-PAS; therefore, embolization treatment was performed.

All CTA scans were performed using the Discovery CT750 HD system (GE Healthcare). During scanning, the left or right median cubital vein was selected for puncture and indwelling to confirm smooth venous access and leakage. Following the preparation, the patient was taken to the CT scan room and was placed in the supine position with the feet advanced. The tube voltage used was 80–100 kVp, which is low, and the tube current was configured using automatic tube current regulation technology. The tube current range was 20–500 mA, and the noise index (NI) was set according to the weight of the child: NI: 11 for 0–16 kg, NI: 13 for 16–35 kg, and NI: 15 for >35 kg. The speed of the tube was 0.5 seconds with a pitch of 0.992:1. The contrast agent used for enhanced scanning was iophorol (320 mgI/mL) (Hengrui Medicine, China), with 1.2–1.6 mL/kg of the contrast agent administered according to the child's weight, and the injection was completed within 15 seconds using a single-cylinder high-pressure syringe. The arterial phase scanning was started at 17 seconds, and the intravenous scanning was performed at 50 seconds. The original image obtained was reduced by 70% weighted adaptive statistical iterative reconstruction-V (GE Healthcare), a practical standard algorithm, without any additional core algorithm. The image slices were reconstructed at 5 and 0.625 mm.

Image evaluation

All images were read by two senior pediatric radiologists. The sites of the lesion and imaging features were recorded, including consolidation, ground-glass opacity (GGO), and reticular opacity. A consensus was reached following discussion.

Statistical analysis

All statistical analyses were performed using Jeffrey's Amazing Statistics Program (version 0.18.1). All continuous variables that conformed to the normal distribution were expressed by mean ± standard deviation ($x \pm s$). The chi-squared test and Fisher's exact test were used to compare CT features between the BA-PAS and non-BA-PAS groups. The level of significance was set at α: 0.05.

Results

Demographics and clinical measures of participants

A total of 74 children with pulmonary hemorrhage were included in this study, including 32 boys and 42 girls, ranging in age from 1 to 12 years, with a mean age of 7.43 ± 3.59 years. Of the 74 children, 36 had diffuse alveolar hemorrhage, including 15 boys and 21 girls. In addition, there were 30 children with BA-PAS, 17 with primary BA-PAS and 13 with secondary BA-PAS, including 12 boys and 18 girls.

Of the 74 children with pulmonary hemorrhage, seven exhibited signs of consolidation in the middle lobe of the right lung, and all of them had BA-PAS, including four cases of primary BA-PAS and three cases of secondary BA-PAS. Consolidation was not observed on chest CT in the other children; BA-PAS in the right middle lobe occurred in 17 cases (10 primary BA-PAS and 7 secondary BA-PAS). BA tortuosity and thickening were observed in all 30 children with BA-PAS.

Correlation analysis

The CT findings were divided into three categories: normal, GGO, and consolidation (Figure 1). All imaging features are presented in Tables 1 and 2. The odds ratios of normal, GGO, and consolidation were 0.533 (P = 0.238), 1.050 (P = 0.918), and 2.250 (P = 0.141), respectively (Table 3).

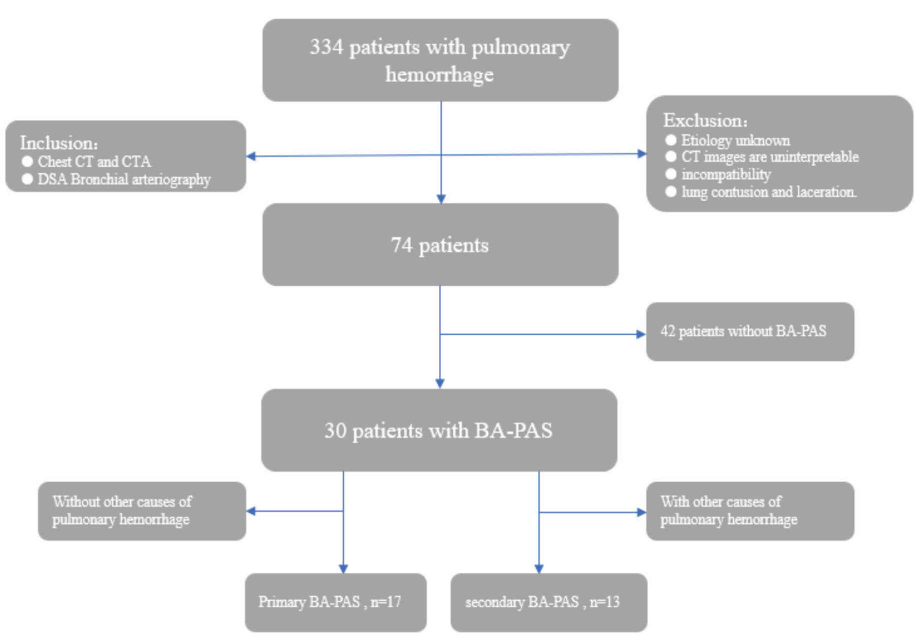

Figure 1. Summary of patient recruitment and exclusions. CT, computed tomography; CTA, computed tomography angiography; DSA, digital subtraction angiography; BA-PAS, bronchial artery–pulmonary artery shunt.

Discussion

The blood in the pulmonary artery is mainly used for air exchange. The BAs supply blood to the lung parenchyma. When abnormalities occur in the lung parenchyma, the blood supply to the BAs increases, leading to lumen thickening and even BA-PAS, which can be primary or secondary. Many diseases can cause BA-PAS, including tumors, diffuse alveolar damage, tumors, and bronchiectasis. Primary BA-PAS intestinal manifestations of recurrent pulmonary hemorrhage are often difficult to distinguish from idiopathic pulmonary hemosiderosis on imaging, and hormone therapy is not effective. Because BA-PAS can be a complication of many diseases, primary BA-PAS is often treated as an exclusive diagnosis. Interventional treatment of BA-PAS is both effective and necessary. As an invasive procedure, angiography needs to be applied with caution. Identification of BA-PAS on CT images is important for both radiologists and pulmonologists.

In this study, of the 74 children with pulmonary hemorrhage, seven exhibited signs of consolidation in the middle lobe of the right lung, and BA-PAS was present in all of them, including four cases of primary BA-PAS and three cases of secondary BA-PAS. Moreover, BA-PAS was observed in 30 children (17 primary BA-PAS cases and 13 secondary BA-PAS cases), and BA tortuosity and thickening were observed in all of them (Figure 2). Regarding primary BA-PAS, the middle lobe and lower lobe of the right lung were involved in 94.1% (16) of the children. Those with a fistula located in the middle lobe of the right lung accounted for 58.8% (10 cases), of which 40.0% (four cases) had consolidation in the middle lobe of the right lung. In this study, 41.2% (seven) of the children with primary BA-PAS exhibited no abnormal changes on chest CT, and 58.8% (10 cases) exhibited abnormal changes in the unilateral lung.

The threshold for BA thickening in adults is 2 mm,[11] and there is no corresponding standard in children, as their arteries are generally compared with the contralateral BAs. In this study, all children with BA-PAS exhibited signs of tortuous and enlarged bronchial arteries on the affected side. Primary BA-PAS lesions are predominantly found in the right lower lobe of the lung, and among children with lesions in this region, approximately 50% exhibit consolidation of the right middle lobe. In this study, while not all children diagnosed with BA-PAS presented with consolidation of the right middle lobe, none of the children classified as Non-BA-PAS showed any signs of such consolidation. Therefore, when there is consolidation in the right middle lobe of the lung in children with pulmonary hemorrhage, bronchial arteriography should be performed promptly to confirm the presence of BA-PAS. If the condition is present, embolization should be performed promptly. In 41.2% of the children with primary BA-PAS, there was no apparent hemorrhage on the chest CT scan. In 58.8% of the children with primary BA-PAS, there was only GGO and consolidation shadows in one lung or lobe. In children with pulmonary hemorrhage who have no abnormalities on lung CT or only unilateral lesions, primary BA-PAS should be strongly suspected. Right middle lobe consolidation in secondary BA-PAS may be related to pulmonary deposition during pulmonary hemorrhage. Non-right middle lobe consolidation, GGO, and normal CT findings were not statistically different between the BA-PAS and non-BA-PAS groups of children with pulmonary hemorrhage.

The accumulation of hemosiderin damages lung tissue and leads to fibrosis.[12] This chronic pathological process leads to dilatation of the BAs and the formation of secondary BA-PAS. The right middle lobe bronchus is susceptible to near or total obstruction because its intraluminal diameter is smaller than that of other lobar bronchi.[13] Furthermore, the fissures of the middle lobe and lingula isolate these segments from collateral ventilation, reducing blood clearance.[14]

Table 1. Data of children with primary BA-PAS

Number	Age (years)	Sex	Location of BA-PAS	CT imaging
1	14	F	RML	RML consolidation
2	13	F	RML	RML, RLL consolidation
3	13	F	RML	Normal
4	12	F	RLL	RLL GGO
5	12	M	RML	Normal
6	11	F	RML	Normal
7	10	M	RLL, RUL	Normal
8	10	F	RLL	Normal
9	8	F	RLL	Right lung multiple GGOs
10	8	F	RUL	Normal
11	7	M	RML	Normal
12	6	F	RML	RML consolidation
13	5	F	RML	RML consolidation; bilateral multiple GGOs
14	3	M	RLL, RUL	Right lung multiple GGOs
15	3	F	RLL	RLL consolidation
16	5	M	RLL	RLL GGO
17	4	M	RUL	Right lung multiple GGOs

BA-PAS, bronchial artery–pulmonary artery shunt; CT, computed tomography; RML, right middle lobe; RLL, right lower lobe; GGO, ground-glass opacity; RUL, right upper lobe; F, female; M, male.

Table 2. Data of children with secondary BA-PAS

Number	Age (years)	Sex	Location of BA-PAS	Clinical diagnosis	CT imaging
1	14	F	RML	DAH	LML GGO
2	12	M	RLL	CF	Bilateral multiple patchy consolidation and GGO
3	9	F	RML, RUL	DAH	Bilateral multiple GGOs
4	9	M	RUL, RLL	IPH	Bilateral multiple GGOs
5	8	M	RLL	Pneumonia	RML consolidation
6	7	F	RML	HT	RML consolidation
7	7	M	RML	Mucoepidermoid carcinoma of main airway	RML consolidation
8	4	F	RML	DAH	Bilateral multiple GGOs
9	3	M	LLL	Pneumonia	LLL consolidation
10	2	F	RUL, RML, RLL	Hemangioma of the right main bronchus	Right lung multiple GGOs
11	2	F	RLL	IPH	Bilateral multiple GGOs
12	2	M	RLL, LLL	IPH	Bilateral multiple GGOs
13	1	M	RLL	Pneumonia	RLL multiple GGOs

BA-PAS, bronchial artery–pulmonary artery shunt; CT, computed tomography; RML, right middle lobe; DAH, diffuse alveolar hemorrhage; LML, left middle lobe; GGO, ground-glass opacity; RUL, right upper lobe; RLL, right lower lobe; IPH, idiopathic pulmonary hemosiderosis; HT, hereditary telangiectasia; LLL, left lower lobe; CF, cystic fibrosis; F, female; M, male.

Table 3. Comparison of the BA-PAS and non-BA-PAS groups of children with hemorrhage

CT findings	BA-PAS (n = 30)			Non-BA-PAS (n = 44)	Odds ratio	*P* value
	Primary (n = 17) (%)	Secondary (n = 13) (%)	Total (%)			
Normal	7 (41.2)	0	7 (23.3)	16 (36.4)	0.533	0.238
GGO	5 (29.4)	9 (69.2)	14 (46.7)	20 (45.4)	1.050	0.918
Consolidation	5 (29.4)	5 (38.5)	10 (33.3)	8 (18.2)	2.250	0.141
RML consolidation	4 (23.6)	3 (23.1)	7 (23.3)	0	-	0.070*

*Fisher's exact test; BA-PAS, bronchial artery–pulmonary artery shunt; CT, computed tomography; GGO, ground-glass opacity; RML, right middle lobe.

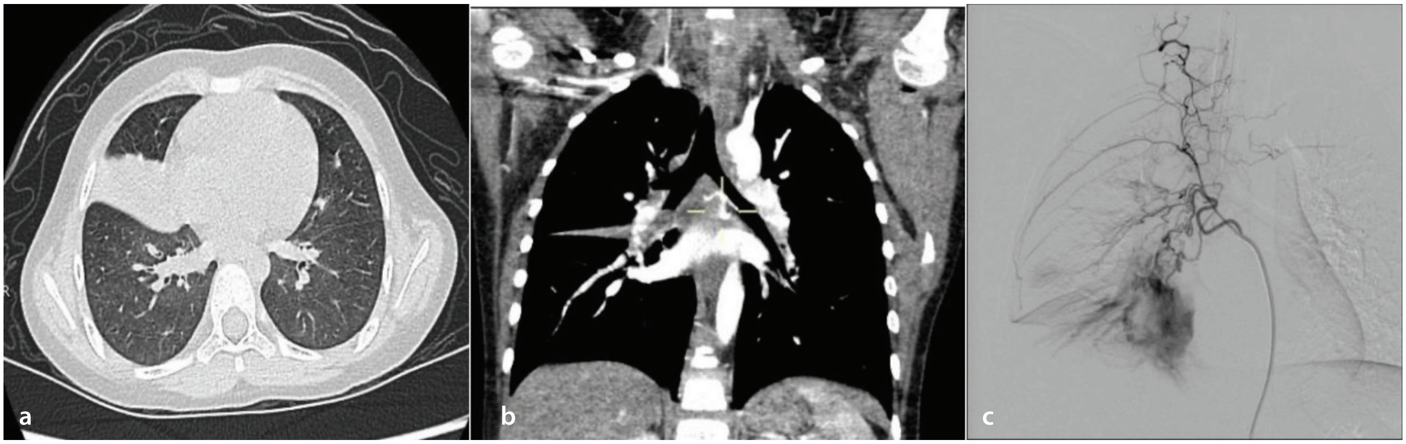

Figure 2. Bronchial artery (BA)–pulmonary artery shunt in a 5-year-old girl: **(a)** computed tomography (CT) scan indicated a patchy dense shadow in the middle lobe of the right lung; **(b)** enhanced CT multiplanar reformation indicated tortuous and thickened BAs; **(c)** digital subtraction angiography–bronchial arteriography indicated pulmonary artery expansion.

The right middle lobe bronchus diverges from the main bronchus at an acute angle, which also leads to obstruction of blood clearance.[15] All of these factors cause blood accumulation in the middle lobe of the right lung, leading to BA-PAS. The cause of consolidation in the middle lobe of the right lung is the same as that of the syndrome in the middle lobe of the right lung. Hemorrhage caused by BA-PAS and blood retention in the middle lobe of the right lung together constitute consolidation in the middle lobe of the right lung on chest CT.

In the authors' experience, some adults with pulmonary hemorrhage and BA-PAS were also observed to have consolidation in the right middle lobe of the lung. Whether there are hemodynamic factors in the middle lobe of the right lung that lead to the formation of BA-PAS needs to be further investigated. The diagnosis of secondary BA-PAS without consolidation in the middle lobe of the right lung is often complex. Close follow-up is required, and the presence of BA-PAS should be suspected if symptoms of pulmonary hemorrhage are not effectively controlled following a period of symptomatic treatment.

In conclusion, BA–PAS can be classified into primary and secondary types. Chest CT images of pulmonary hemorrhage in children are varied and may even be negative. Primary BA-PAS typically manifests as GGO or consolidation in one side of the lung. Regardless of type, there is consolidation in the right middle lobe of the lung on chest CT in children with pulmonary hemorrhage. At this time, bronchial arteriography should be actively performed to confirm the diagnosis.

Footnotes

Conflict of interest disclosure

The authors declared no conflicts of interest.

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