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Hemoptysis requiring bronchial artery embolization in pulmonary arterial hypertension: one center experience

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Abstract

Introduction—Hemoptysis is an uncommon complication in patients with pulmonary arterial hypertension (PAH). Although the mechanism of hemoptysis is unknown, treatment with bronchial artery embolization (BAE) is proposed as a safe and reliable method of treatment. We report Baylor PH Center experience in treating PAH patients presenting with acute hemoptysis that required multiple BAEs.

Results—Three female and one male PAH patients ages 45 ± 9 years (mean \pm SD) presented with acute hemoptysis. Right ventricular systolic pressure and cardiac index at the time of first episode of hemoptysis was 85 ± 17 mm Hg and 2.7 ± 0.7 L/min/m² respectively. Two of the four patients had recurrent episode of hemoptysis requiring multiple BAEs. All four were on intravenous prostacyclin analogue. None were receiving warfarin or endothelin receptor antagonist at the time of the episode. During each episode of hemoptysis INR was 1.09 ± 0.11 units and platelet count was $124,000 \pm 47,000$ per microliter. Each episode of hemoptysis was acutely terminated with BAE. In majority of cases, patients had multiple aberrant bronchial arteries embolized and an average of 2.3 arteries was embolized per session (1–4 embolized arteries). Each BAE was performed utilizing polyvinyl alcohol particles ranging from 250–500 microns. There were no reported complications of the 14 BAE procedures performed.

Conclusion—Although the incidence of hemoptysis is unknown and likely underreported, we report our experience where recurrent hemoptysis was treated with multiple BAE procedures. This report emphasizes the efficacy and safety of BAE in terminating episodes of recurrent hemoptysis in patients with severe PAH.

Keywords

right ventricular systolic pressure; echocardiogram; right heart catheterization

Introduction

Hemoptysis can be a serious complication for patients with pulmonary arterial hypertension (PAH) [1]. It has been reported as a terminal event related in PAH. The mechanism of

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hemoptysis in PAH remains unknown. Treatment with bronchial artery embolization (BAE) is proposed to be a safe and reliable method of terminating hemoptysis. However, there is paucity of data in treating patients with severe PAH with BAE, some of whom may require repeated BAE procedures. We report Baylor PH Center experience in treating PAH patients presenting with hemoptysis and requiring multiple BAE procedures.

Materials and Methods

We retrospectively reviewed charts of PAH patients who presented with hemoptysis between 2004 and 2009. Patient demographic and hemodynamic information at time of diagnosis is summarized in Tables 1 and 2. The episodes of hemoptysis were all treated and terminated with BAE performed by an interventional radiologist. Bronchial artery embolizations were performed by utilizing polyvinyl alcohol (PVA) particles as the embolizing agent. The known therapies for each patient and coagulation studies are given in Table 3. Hemodynamic data and the bronchial arteries embolized are summarized in Table 4.

Results

Patient A

34 year old white male who was diagnosed with PAH at the age of 26. He initially presented with exertional dyspnea and chest pain. His primary care physician ordered an echocardiogram, which revealed elevated pulmonary systolic pressure and he was referred to the Baylor PH center and underwent right heart catheterization confirming the diagnosis of PAH. The patient was started on intravenous epoprostenol due to severe symptoms and poor functional class. Digoxin and diuretics were added. He initially presented with hemoptysis of approximately 5 cc of bright red blood 3 times a day for four days. This episode occurred approximately 2 years following diagnosis of PAH. Thoracic aortogram revealed hyperemia of the bronchial arteries, with subsequent selective embolization of two right bronchial arteries and one left bronchial artery with PVA (250–350 microns) (Figure 1). Hemoptysis resolved following embolization and the patient stayed asymptomatic for approximately four months. However, at that time, he once again presented with hemoptysis and a markedly enlarged right bronchial artery was embolized with successful termination of symptoms. The patient continued to have recurrent episodes of hemoptysis, which have always been successfully terminated with BAE. He has had a total of 21 bronchial arteries embolized for 9 individual episodes of hemoptysis over a period of 5 years.

Patient B

54 year old white female who was diagnosed with PAH at the age of 52. She initially had multiple episodes of syncope and was found to have elevated pulmonary artery systolic pressures on an echocardiogram. Right heart catheterization was performed to confirm the diagnosis. She was started on intravenous epoprostenol in addition to diuretics and digoxin. The patient presented to the clinic approximately 7 months after the diagnosis with complaints of incessant cough and approximately 100 cc of acute hemoptysis. The patient was admitted and a thoracic aortogram revealed a small right bronchial artery and

collateralized left bronchial artery. These bronchial arteries were selectively embolized with PVA (250–350 microns). The hemoptysis symptoms resolved following embolization. The patient subsequently expired from presumed cardiac arrhythmia approximately three year following PAH diagnosis.

Patient C

47 year old white female with history of ventricular septal defect and Eisenmenger's syndrome with severe PAH. She has marked limitation of physical activity due to dyspnea and chest pain. She had been maintained on intravenous treprostinil, diuretics and digoxin when she presented to the emergency room with 60 cc of bright red hemoptysis. The patient was taken to the interventional radiology suite and thoracic aortogram revealed two enlarged right and two enlarged left bronchial arteries. All four were embolized with PVA (250–355 microns) achieving adequate occlusion. The patient recovered and has had no further episodes of hemoptysis.

Patient D

46 year old African American female with history of rheumatoid arthritis and PAH diagnosed at the age of 36 who was on intravenous epoprostenol since her diagnosis. This patient had two separate episodes of hemoptysis both treated successfully with bronchial artery embolization. Two bronchial arteries were embolized with PVA (355–500 microns) following the first episode of hemoptysis. The second episode occurred approximately 3 months after the first episode and one bronchial artery was embolized with PVA (250–355 microns). No further hemoptysis has occurred since embolization, for approximately 4 years.

Discussion

Recurrent hemoptysis can be managed by repeating BAE. In our experience, four patients with severe PAH on intravenous prostacyclin therapy presented with hemoptysis (Table 2). Recurrent hemoptysis occurred in 2 of these patients requiring repeated BAE. One patient had repeated episodes of hemoptysis that required bronchial artery embolization a total of 9 times. Prostacyclin have been reported to reduce platelet counts[2]. However, marked thrombocytopenia or abnormal coagulation profile was not noted during each hemoptysis episode. Each of these patients received BAE with no complications from the procedure and resulted in resolution of the hemoptysis. Hemoptysis reoccurred in two of the four patients requiring repeat procedures that did not lead to any complications.

Epidemiology of PAH

Although the prevalence and incidence of patients with IPAH is unknown in the United States, the REVEAL registry may provide further insight [3, 4]. In France, a national registry was established, and the prevalence and incidence of PAH in France were estimated to be 15.0 cases per million of adults and 2.4 cases per million of adults per year [5]. These estimates were consistent with recently published data from a Swiss national registry which reported prevalence of PAH as 15.5 patients per million adults and an incidence of 3.5 patients per million adults per year [6].

Despite the availability of multiple registries of patients with PAH, the true incidence of clinically significant hemoptysis is unknown. A review of the literature by Reesink, et. al. revealed only 4 reported cases of patients with PAH without chronic thromboembolic pulmonary hypertension in 1844 patients presenting with hemoptysis [7]. Although it appears that hemoptysis may be a rare symptom in patients with PAH, it is likely that there is significant underreporting in the literature [7].

Hemoptysis

The underlying physiologic mechanism of hemoptysis in patients with PAH has yet to be defined. Hemoptysis in patients with pulmonary hypertension due to chronic thromboembolic disease (CTED) has been reported [7, 8]. The reason for hemoptysis in patients with CTED is likely secondary to bronchial artery hypertrophy and extensive collateral angiogenesis [9]. Remy-Jardin et. al. compared the systemic collateral supply of 22 patients with CTED and 14 patients with PAH using multisection spiral CT angiography [10]. The frequency of enlarged bronchial and non-bronchial systemic arteries was much lower in the PAH patients as compared to CTED patients [10]. Therefore, the pathologic mechanism may actually differ between patients with PAH and those with pulmonary hypertension secondary to CTED.

Bronchial Artery Embolization

There have been no randomized trials performed to identify the best treatment for PAH patients with hemoptysis. Multiple case series report successful termination of acute hemoptysis with use of BAE. [1, 7, 11]. Bronchial artery embolization is performed after a descending thoracic aortogram. The preliminary aortogram is used to determine the number and origin of bronchial arteries [12]. Angiographic findings in patients with hemoptysis may include hypertrophic and tortuous bronchial arteries, hypervascularity, neovascularity and rarely, extravasation of contrast medium [13, 14]. Differing techniques with modern embolizing agents have been described [12, 15–19]. Bronchial artery embolization appears to be highly successful in acute termination of hemoptysis. However, it is not uncommon for a patient to have recurrent hemoptysis requiring re-embolization [1, 20–22]. Recurrence rates of hemoptysis in PAH patients treated with BAE remains unknown. Complications of BAE appear to be rare and include non-target embolization, subintimal dissection and arterial perforation [1, 23]. Bronchial artery embolization is a safe and effective therapy for hemoptysis due to multiple etiologies, including PAH.

Conclusion

Hemoptysis is an uncommon symptom in patients with PAH, although incidence is unknown and likely underreported. There have been no randomized trials that explore optimal therapies, although BAE appears to be the best therapy for this condition. This case series emphasizes the efficacy and safety of BAE in terminating recurrent episodes of hemoptysis in patients with PAH.

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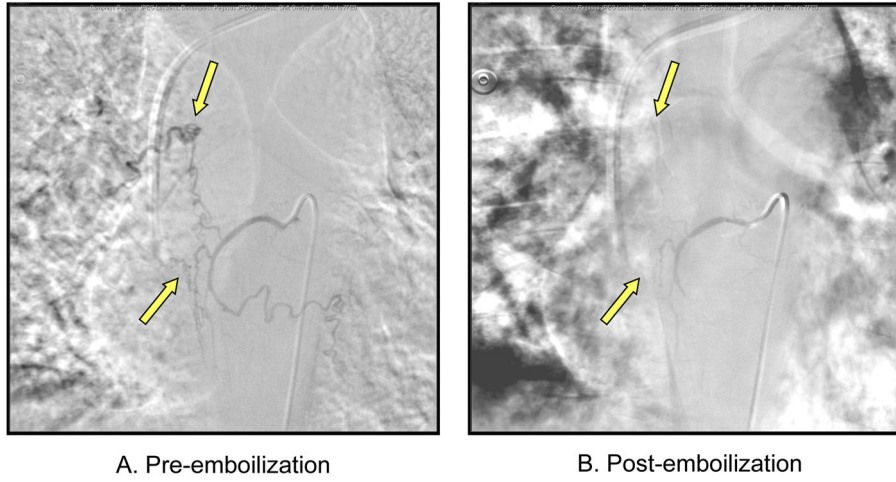


Figure 1.
Bronchial Artery Angiogram showing hypertrophied bronchial arteries

Table 1

Demographics characteristics of the PAH patients.

	Age at diagnosis (yrs)	Age at first hemoptysis (yrs)	Race	Gender	Etiology of PAH	BMI
Patient A	26	29	Caucasian	M	Idiopathic	33
Patient B	52	53	Caucasian	F	Idiopathic	49
Patient C	NA	46	Caucasian	F	CHD	24
Patient D	36	42	Africo-American	F	CVD	31

NA, not available; CHD, congenital heart disease; CVD, collagen vascular disease

Table 2

Hemodynamics at the time of PAH Diagnosis.

	PA (S/D) mm Hg	mean PA	CO (L/min)	CI (L/min/m ²)	RAP (mm Hg)
Patient A	112/58	76	1.9	1.0	10
Patient B	120/40	66	5.3	2.3	33
Patient C	115/42	66	4.2	2.3	8
Patient D	80/40	53	2.9	1.3	14

PA (S/D), pulmonary artery systolic/diastolic pressure; PA, pulmonary artery; CO, cardiac output; CI, cardiac index; RAP, right atrial pressure.

Table 3

Medications and laboratory values for each patient at the time of hemoptysis

Patient	Episode	ERA	PDE-5I	Digoxin	Warfarin	Platelets (microliter)	INR (unit)	BUN (mg/dL)	Creatinine (mg/dL)
A	1	No	No	Yes	No	72	1	16	1.3
	2	No	No	Yes	No	63	1.1	17	1.4
	3	No	No	Yes	No	61	1.1	14	1
	4	No	Sildenafil	Yes	No	107	1.1	15	1.2
	5	No	Sildenafil	Yes	No	90	1	17	1.1
	6	No	Sildenafil	Yes	No	106	1	16	1.2
	7	No	Sildenafil	Yes	No	151	1.1	15	0.8
	8	No	Sildenafil	Yes	No	129	1.1	13	1.1
	9	No	Sildenafil	Yes	No	118	1.1	17	0.9
	1	No	No	Yes	No	172	0.9	22	1
C	1	No	No	Yes	No	165	1.1	15	1
D	1	No	No	Yes	No	161	1.3	11	1
	2	No	No	Yes	No	214	1.3	10	0.9

ERA, endothelin receptor antagonist; PDE5-I, phosphodiesterase 5-inhibitor.

Table 4

Hemodynamic data at each hemoptysis episode

Patient	Episode	PG	Dose (ng/kg/min)	RVSP (mm Hg)	CO (L/min)	CI (L/min/m ²)	RAP (mmHg)	PE	Bronchial Artery Embolized
A	1	Epo	17	98	5.53	2.74	10	No	Right X2, Left
	2	Epo	17						Right
	3	Epo	15.5	92.5	4.4	2.2	7.5	No	Right, Left
	4	Epo	30	85.5	3.83	1.8	7.5	No	Right X2, Common
	5	Epo	30	82.5	3.83	1.8	7.5	No	Right, Left
	6	Epo	30	115.5	3.37	1.6	17.5	No	Right, Left
	7	Trep	48						Right, Common
	8	Trep	48	112.5	ND	ND	12.5	No	Right X2, Left
	9	Trep	48	ND	ND	ND	ND	ND	Right, Left, Common
B	1	Epo	33	74.5	5.36	2.35	7.5	No	Right, Left
	1	Trep	143	102	6.1	3.77	10	Very Small	Right X2, Left X2
D	1	Epo	118	66	4.49	2.22	5	No	Right, Left
	2	Epo	118	74.5	5.8	2.9	7.5	Very Small	Right

Epo, epoprostenol; Trep, treprostimil; RVSP, right ventricular systolic pressure; CO, cardiac output; CI, cardiac index; RAP, right atrial pressure; PE, pericardial effusion.