

CASE REPORT

N-Butyl Cyanoacrylate Embolization of a Peripheral Pulmonary Artery Aneurysm in a Patient with Patent Ductus Arteriosus

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Abstract: N-Butyl cyanoacrylate embolization of peripheral pulmonary artery aneurysms has rarely been performed. This report presents a young woman with a history of patent ductus arteriosus and massive hemoptysis secondary to pulmonary artery aneurysm, successfully managed by embolotherapy using n-Butyl cyanoacrylate. This is the first report of n-Butyl cyanoacrylate embolization treatment of a bleeding pulmonary artery aneurysm in the setting of patent ductus arteriosus. The pertinent literature has been reviewed and the clinical background and the procedure have been explained.

Keywords: N-Butyl cyanoacrylate, pulmonary artery aneurysm, pulmonary artery embolization, patent ductus arteriosus, hemoptysis, embolotherapy.

1. INTRODUCTION

Massive hemoptysis can be a fatal condition and usually originates from bronchial arteries. However, in about 10 percent of the cases, pulmonary arteries are responsible for the condition and in some cases, both bronchial and pulmonary arterial systems are involved [1, 2].

Pulmonary artery aneurysms (PAAs) and pulmonary artery pseudoaneurysms (PASAs) are rare and commonly associated with pulmonary hypertension (PH) [2, 3]. Peripheral PAAs and PASAs are rarer and involve pulmonary arterial branches distal to main pulmonary arteries [3]. Infections (with the TB as the hallmark), pulmonary hypertension (PH) and a congenital or acquired defect in the vessels wall (vasculitids) are main etiologies of the disorder [3, 4]. Although surgery has been traditional management of the abnormality, endovascular intervention is increasingly used by considering risks of surgery [1-5].

This is the first report of the endovascular treatment of a peripheral PASA with N-Butyl cyanoacrylate (NBCA) in a patient with patent ductus arteriosus (PDA).

2. CASE PRESENTATION

A 21 Year-old girl was brought to the emergency department because of hemoptysis. Multiple witnessed episodes of hemoptysis were noted, each with more than a

cup of blood expectorated. Serum hemoglobin level was 5mg/100ml with O₂ saturation of 80 percent. This was preceded by episodes of intermittent mild hemoptysis in the prior three weeks. She carried an established diagnosis of PDA and awaiting surgical correction. Echocardiographic evaluation revealed severe pulmonary hypertension. Spiral contrast enhanced chest computed tomography showed possible focal contrast extravasation in the base of the left lung (Fig. 1).

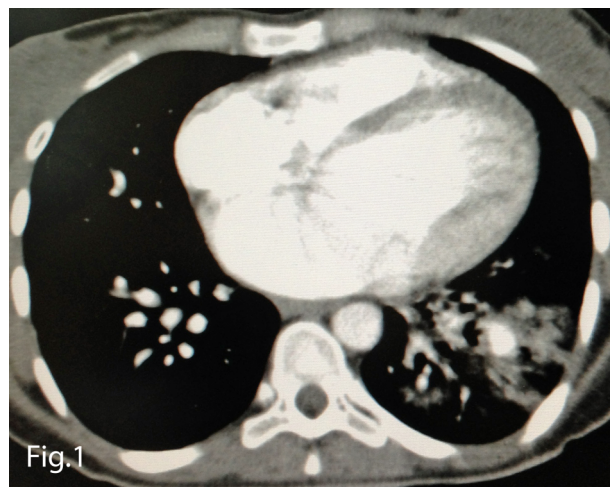


Fig. (1). Computerized Tomography scan of the chest demonstrating focal contrast accumulation in the left lower lobe with surrounding alveolar densities. (A higher resolution / colour version of this figure is available in the electronic copy of the article).

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With the possibility of peripheral PASA, she was deemed to be a high-risk surgical candidate due to severe pulmonary hypertension. An endovascular approach was considered as the first line of treatment.

Selective left pulmonary angiography *via* right femoral vein access confirmed the presence of a nearly 2 centimeter pseudoaneurysm arising from descending branch of the left pulmonary artery (Fig. 2).



Fig. (2). Digital subtraction selective angiography of the left lower trunk pulmonary artery depicting an about 2cm somewhat irregular shape aneurysm. (A higher resolution / colour version of this figure is available in the electronic copy of the article).

Embolic coils or other suitable embolic agents were unavailable in our hospital at the time of angiography with the exception of NBCA (Nycomed-Ingenor, Paris, France). We elected to treat the patient given the gravity of her clinical condition. *Via* a 5-Fr base catheter in the feeding artery, the pseudoaneurysm sac was subselectively cannulated with a Vasco 1.8 Fr microcatheter (Balt Extrusion, Montmorency, France). Considering the inner dead volume of the microcatheter, approximately 2 milliliters of 1:1 mixture of NBCA and Lipiodol (Guerbet, Aulnay-sous-Bios, France) was used to obliterate the lumen and neck of the pseudoaneurysm. After injection of approximately 1.5 cc of the liquid embolic mixture, severe increase in resistance was noted and the microcatheter was found to be occluded, which was removed using a quick jerky motion. Repeat angiogram *via* the base catheter revealed complete obliteration of the sac with preservation of normal flow in the segmental branches distal to the lesion (Fig. 3). The patient underwent a successful surgical ligation of the PDA three months later with no other episode of hemoptysis in the interval.



Fig. (3). Repeat digital subtraction angiography of the left lower trunk pulmonary artery revealing obliteration of the aneurysm. (A higher resolution / colour version of this figure is available in the electronic copy of the article).

3. DISCUSSION

PAA and PASA are rare disorders which can propose a treatment dilemma, particularly in patients with comorbidities. In a report by Deterring and Clagett, only 8 PAAs were found in a review of 109,571 necropsies (about 0.007 percent) [1]. Peripheral type of the disorder which include those involving pulmonary arterial branches distal to the main pulmonary arteries is even rarer. In a series of 111 cases of PAAs, only 22 (about 20 percent) were found in the peripheral portion [1, 3]. In a recent report of 76 cases with hemoptysis, 8 (about 10 percent) were due to peripheral PAA [4]. Infections (especially TB), pulmonary hypertension (usually in the setting of congenital heart disease), vasculitides (especially Behçet's disease) and trauma (including iatrogenic cases) are among the etiologies [1-4].

Contribution of pulmonary hypertension is a less reported etiology in patients with peripheral PAA compared with proximal PAA [1]. In a necropsy review of 111 cases of PAAs, pulmonary hypertension due to cardiac malformation was present in 66 percent with PDA as the most common single lesion in 22 percent of the cases [3]. Both of the two reported cases of peripheral PAA by Sacuma *et al.*, suffered from idiopathic pulmonary hypertension. On the other hand, none of the 6 cases of PAAs and PASAs reported by Krokidis *et al.* [2], 24 cases of peripheral PAPAs reported by Shin *et al.* [5] and eight PAAs in the study by Sbano *et al.* [4] were associated with pulmonary hypertension.

In the cases of peripheral PAAs surgery (lobectomy and/or pneumonectomy) has been the traditional method of treatment with a high mortality and morbidity [1, 5]. Increasing number of endovascular interventions are being

reported as a less invasive technique, especially considering increased risk of surgery in the cases associated with pulmonary hypertension [1, 5].

When the percutaneous route is chosen for treatment of massive hemoptysis, there is always a question of whether to embolize pulmonary or systemic arteries [2, 5]. Before the advent of computed tomography angiography (CTA), embolotherapy of bronchial arteries was the first line management of massive hemoptysis [5]. With the use of CT, as in our case, other pulmonary vascular disorders are detected necessitating a different plan of treatment [5]. Of the 6 cases managed by Krokidis *et al.*, embolization of the pulmonary arteries was adequate for control of the hemoptysis [2]. Of the 11 patients treated by Remy *et al.* for massive hemoptysis attributed to PAAs, five patients died after isolated embolization of bronchial arteries with no treatment of the pulmonary side [6]. They and other authors have suggested embolization of pulmonary arteries is enough and should be the first procedure in cases with known involvement of the pulmonary tree [2, 3, 6]. On the other hand, Shin *et al.* performed CTA and both bronchial and pulmonary angiography on 24 patients with hemoptysis due to peripheral PASAs. Fifteen lesions were detected by non-selective or selective pulmonary artery angiogram and managed successfully by embolization of both bronchial and pulmonary arteries. Of the remainder, five cases were detected on the bronchial artery or systemic arterial catheterization but not at selective pulmonary angiography and four cases were demonstrated on pulmonary CT alone but not at pulmonary or bronchial catheterization. Embolization alone of the bronchial and non-bronchial systemic collateral arteries was performed for these patients. Hemoptysis was not controlled in 3 of the nine patients; of which two underwent percutaneous injection therapy and surgical resection was performed for one patient [5].

Several approaches are available for endovascular treatment of patients with peripheral PAAs and PASAs [1, 2, 5]. Metallic coils are the preferred option for endovascular management of PAAs but may not be cost effective, depending on the size of treatment [2]. In addition, in PASAs with fragile walls the use of coils bears a potential risk of rupture of the lesion [7, 8]. In such cases, especially when the pulmonary artery feeding the pseudoaneurysm is irregular, proximal and distal occlusion of the irregular artery (front door- back door technique) has been considered as the optimal procedure by some authors [4, 7]. Most authors have used coils or microcoils with the intent to preserve the originating pulmonary artery [2]. Migration is one of the concerning factors, especially in the cases with wide aneurysm neck. However, the "caging technique" or "stent assisted coiling" has been used in such scenarios [2].

A few reports of the use of liquid embolic agents (such as alcohol and NBCA) for endovascular treatment of peripheral PAAs and pseudo-aneurysms are available in the literature [2, 8-12]. NBCA is widely used as a liquid embolic material to treat brain and peripheral arteriovenous malformations, GI bleeding, varicoceles, and endoleaks [7]. It has also been used in a few patients with hemoptysis due to ruptured PAAs in Behçet's disease with the "bubble technique" [8, 11]. Cantasdemir *et al.* used NBCA instead of coils by

considering the potential risk of rupture of the PSAs and cost of the coils [8]. It is a safe agent in experienced hands but with the risks of systemic embolization and catheter adhesion as the major drawbacks [11]. Recanalization has been reported, especially when only partial embolization was done [11]. Percutaneous injection of Thrombin and NBCA under fluoroscopic or ultrasound guidance has also been used when the feeding artery of the pseudoaneurysm cannot be identified at pulmonary angiography [12].

CONCLUSION

Different endovascular techniques have been proposed for the treatment of peripheral pulmonary artery aneurysm and peripheral pulmonary artery pseudoaneurysm each with unique drawbacks and benefits. This case report which includes a few previously reported cases in the literature shows that NBCA could be safely used in the emergency liquid embolotherapy of these patients with massive hemoptysis.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

Not applicable.

HUMAN AND ANIMAL RIGHTS

No animals/humans were used for studies that are base of this research.

CONSENT FOR PUBLICATION

Not applicable.

STANDARD OF REPORTING

CARE guidelines and methodology were followed in this study.

AVAILABILITY OF DATA AND MATERIALS

Not applicable.

FUNDING

None.

CONFLICT OF INTEREST

The authors declare no conflict of interest, financial or otherwise.

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Declared none.

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