Original Article

# Endovascular glue embolization for control of massive hemoptysis caused by peripheral pulmonary artery pseudoaneurysms: Report of 7 cases

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## ABSTRACT

**Purpose:** Pulmonary artery pseudoaneurysms are a rare cause of massive hemoptysis and need to be considered as a differential with prompt recognition preventing mortality from life-threatening hemorrhage. We report the clinical details and imaging findings for a series of patients presenting with massive haemoptysis due to peripheral pulmonary artery pseudoaneurysm, managed by endovascular glue embolization. **Methods:** Hospital based retrospective study wherein records and image database of patients presenting with massive hemoptysis between January 2014 and March 2016 were studied. The immediate technical success, reintervention rate, and recurrence of massive hemoptysis were recorded. **Results:** Among patients with massive hemoptysis who underwent endovascular management in our department, seven patients fulfilled the inclusion criteria. The mean age was 54.3 years; mean lesion diameter was 10.8 mm (range 6–14 mm); underlying pathology being infective (tuberculosis) in all cases ($n = 7$). All lesions were treated with endovascular glue embolization. The technical success was 100%. Mean follow-up was 11.7 months. There was no case of recurrence of massive hemoptysis. There were no major complications with a single case of minor complication (in the form of chest pain and discomfort) which resolved with medical management. **Conclusion:** The endovascular approach using glue (N-butyl cyanoacrylate) is a minimally invasive and technically feasible, effective technique for emergent management of pulmonary artery pseudoaneurysms presenting as massive hemoptysis.

**KEY WORDS:** Pulmonary artery pseudoaneurysm, endovascular glue embolisation, hemoptysis

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## INTRODUCTION

Massive hemoptysis is defined as expectoration of >300 ml of blood from the bronchial tree within 24 h. It is an emergent condition and is associated with mortality rate of >50%. The majority of cases have bronchial arteries as the source of hemoptysis, however, in <10%, it results from the pulmonary arteries.[1] Rarely both the systems may be involved.

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Pulmonary artery pseudoaneurysms are a rare cause of hemoptysis with the etiology being diverse including trauma, iatrogenic (Swan-Ganz catheter placement), infections such as tuberculosis, aspergillosis and necrotizing pneumonias, and necrotic tumor. The treatment options include surgical management, percutaneous, and endovascular embolization. Surgery is associated with high morbidity and mortality,



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especially when done in the emergent setting; however, endovascular transcatheter embolization offers the possibility of improved safety and efficacy while being less invasive. Endovascular catheter-directed treatment techniques include embolization using coils, use of closure device, or embolization using liquid embolic agents. An endovascular approach using glue (N-butyl cyanoacrylate) is commonly described as embolization technique for visceral artery pseudoaneurysms. We report the seven cases of peripheral pulmonary artery pseudoaneurysms associated with pulmonary tuberculosis in which massive hemoptysis was successfully controlled by intra-aneurysmal glue injection.

## METHODS

The hospital-based retrospective study was done after taking approval from clinical audit department of our hospital. Institutional review board approved this study for retrospective review of patient's record and images. Informed consent was waived by our Institutional review board because we included only those patients who had previously provided authorization for the use of their medical records for research. We evaluated the records of patients presenting to emergency department with massive hemoptysis for the period between January 2014 and March 2016. We report a total of seven patients who had peripheral pulmonary artery pseudoaneurysm on computed tomography (CT) angiography and underwent endovascular intra-aneurysmal glue injection for control of hemoptysis. Peripheral pulmonary artery aneurysms are defined as those which arise beyond the second-order branch of the pulmonary artery. Observations were tabulated under the headings of age, gender, disease laterality, underlying etiology, angiography findings, technical success, complications, and follow-up. Primary end-points were immediate technical success and reintervention rate; secondary end-point was relapse of massive hemoptysis.

All seven patients having pulmonary artery pseudoaneurysms (PAP) were males with mean age of 54.3 years (range of 35–77 years). In all patients, the procedure was done in emergent setting. In seven patients, there were a total seven PAP detected on CT scan. The mean lesion diameter was 10.8 mm (range 6–14 mm). The lesions were detected in apical segmental artery of the right upper lobe ($n$ = 1), anterior segmental artery of the left upper lobe ($n$ = 1), right lower lobe superior segmental artery ($n$ = 1), left lower lobe superior segmental artery ($n$ = 2), and distal branches of left lower lobe anteromedial segmental artery ($n$ = 2). All the lesions were saccular ($n$ = 7) in nature.

All seven patients of PAP had either recent or old underlying infection due to pulmonary tuberculosis diagnosed by sputum acid–fast bacillus staining or Genexpert (molecular DNA testing for tuberculosis bacteria). The observations have been summarized in Table 1.

After percutaneous catheterization of the right femoral vein using the Seldinger technique, an 11 cm, 6-Fr sheath (Avanti Sheath Introducer, Cordis, Ireland) was introduced into the right femoral vein. Selective pulmonary angiography was done using manual contrast injection based on anatomical details provided with CT pulmonary angiography. 6 Fr Judkin's right guiding catheter (Cordis, Milpitas, California, USA) over a 0.035" guidewire (glide wire, Terumo, New Jersey, USA) was advanced through the inferior vena cava and the right atrium into pulmonary circulation followed by selective catheterization of the culprit pulmonary artery branch with a 2.7 Fr progreat catheter (Terumo, New Jersey, USA). The tip of catheter was passed through the neck of aneurysm, and a selective contrast injection was done to confirm the position of the catheter. About 50% cyanoacrylate glue (N-butyl cyanoacrylate mixed with lipiodol in equal concentrations) was injected selectively into the pseudoaneurysm under fluoroscopic guidance till complete obliteration of aneurysmal cavity. The strategy of glue injection was to fill the aneurysm sac, and in process if there was occlusion of diseased branch of pulmonary artery supplying the pseudoaneurysm, it was considered acceptable. After the completion of procedure, an intraprocedure angiogram was taken to demonstrate

**Table 1: Clinical details, angiography findings, technical success, complications and follow-up of patients presenting with massive hemoptysis due to pulmonary artery pseudoaneurysm and managed using intra-aneurysmal glue injection**

| Age (years) | Gender | Disease laterality | Etiology | Angiographic findings | Technical success | Complications | Follow-up |
|---|---|---|---|---|---|---|---|
| 49 | Male | Right upper lobe | TB | 6.5 mm × 5.7 mm sized pseudoaneurysm in apical segment of RUL | Yes | None | 16 months |
| 51 | Male | Right lower lobe | TB | 11 mm × 9 mm sized PAP in superior segment of right lower lobe | Yes | None | 9 months |
| 56 | Male | Left lower lobe | TB | 11 mm × 9 mm in PAP in superior segment of left lower lobe | Yes | Minor | 12 months |
| 77 | Male | Lingula, left lower lobe, right | TB | 15 mm × 13 mm sized PAP in anteromedial basal segment of the left lower lobe | Yes | None | 5 months |
| 35 | Male | Left >right | TB | 13 mm × 10 mm sized PAP in anteromedial basal segment of left lower lobe | Yes | None | 9 months |
| 40 | Male | Left lower lobe | TB | 18 mm × 10 mm sized PAP in superior segment of left lower lobe | Yes | None | 25 months |
| 72 | Male | Left upper lobe | TB | 11 mm × 10 mm sized PAP in the anterior segment of the left upper lobe | Yes | None | 6 months |

TB: Tuberculosis, PAP: Pulmonary artery pseudoaneurysm, RUL: Right upper lobe

the complete embolization of pseudoaneurysm. In cases with simultaneous presence of dilated, tortuous and hypertrophied bronchial arteries diagnosed on CT angiography; a femoral arterial approach was also taken, and they were successfully embolized using polyvinyl alcohol particles (500–700 um PVA, Cook Medical, Denmark) so as to prevent the recurrence of hemoptysis [Figures 1-3 show the representative cases, preembolization image, technique, and postembolization angiograms].

Technical success was defined as complete embolization of aneurysmal sac with no residual contrast opacification on postembolization intraprocedural angiography and cessation of hemoptysis. Procedure-related complications were characterized into major and minor according to international standards (Society of Interventional Radiology classification for complications by outcome).

## RESULTS

The technical success rate was 100% with all patients showing successful embolization of pseudoaneurysm and cessation of massive hemoptysis. All patients were discharged after 3 days postprocedure and were regularly followed up in outpatient department of hospital with the mean follow-up of 11.7 months. There were no major complications related to the procedure. One patient had minor complication in the form of chest pain and discomfort which resolved with conservative management in 2 days.

## DISCUSSION

Pulmonary artery aneurysms and pseudoaneurysms are rare causes of massive hemoptysis.[1,2] Pulmonary artery aneurysms commonly originate from main pulmonary artery with few cases described in peripheral pulmonary arteries as well. Pseudoaneurysms are false aneurysms, which consist of a single layer of fibrous tissue surrounding a sac of turbulent blood flow, usually surrounded by hematoma.[3] Because they lack an adventitial wall, they are more prone to enlargement and rupture. The etiology is varied and is usually due to arterial wall disruption secondary to trauma, iatrogenic interventions, infection, vasculitides and erosion by a necrotic tumor.[2,4,5] In our study, all seven cases had pulmonary tuberculosis as the etiology. Rasmussen aneurysms occur adjacent or within a tubercular cavity and are usually distributed peripherally beyond the branches of main pulmonary artery. The infection leads to destruction of the vessel wall and replacement with granulomatous tissue from outer to inner wall.

CT angiography with evaluation of pulmonary and bronchial arteries is the first-line investigation to suggest source of bleeding. It gives information about location,

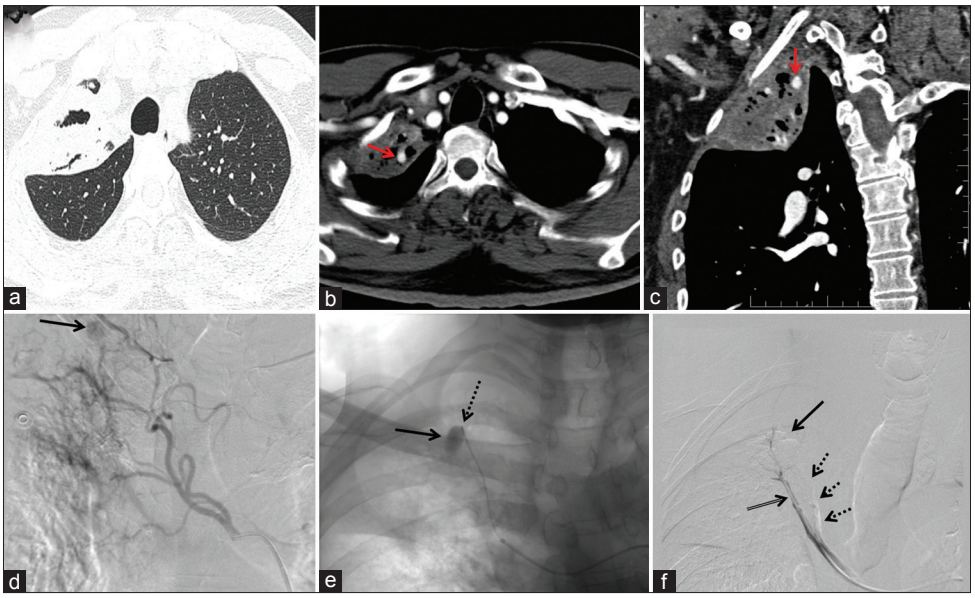


**Figure 1:** Axial section of high-resolution computed tomography chest (a) in lung window showing subsegmental areas of cavitatory consolidation in the right upper lobe. Axial and coronal reformatted sections of contrast enhanced computed tomography chest (b and c) showing 6.5 mm × 5.7 mm sized pulmonary artery pseudoaneurysm (red arrow) noted arising from apical segmental branch of the right ascending pulmonary artery. Anteroposterior projection of catheter angiography (d) with selective cannulation of the right intercostobronchial trunk showing dilated, tortuous, hypertrophied bronchial vessels with blush in the right lung field and opacification of pseudoaneurysm (black arrow) and right ascending pulmonary artery due to bronchopulmonary shunt. The intercostobrachial trunk was embolized using polyvinyl alcohol particles, and the information was used for selective cannulation of the right ascending pulmonary artery pseudoaneurysm. Anteroposterior projection of catheter pulmonary angiography (e) selective injection of contrast after cannulation of pseudoaneurysm (dashed black arrow showing the tip of microcatheter in pseudoaneurysm) showing opacification of pseudoaneurysm (black arrow). Anteroposterior projection digital subtraction angiography image (f) showing glue embolization of pseudoaneurysm (black arrow) and its feeding artery (dashed black arrow) with normal opacification of adjacent branch (double black arrow)

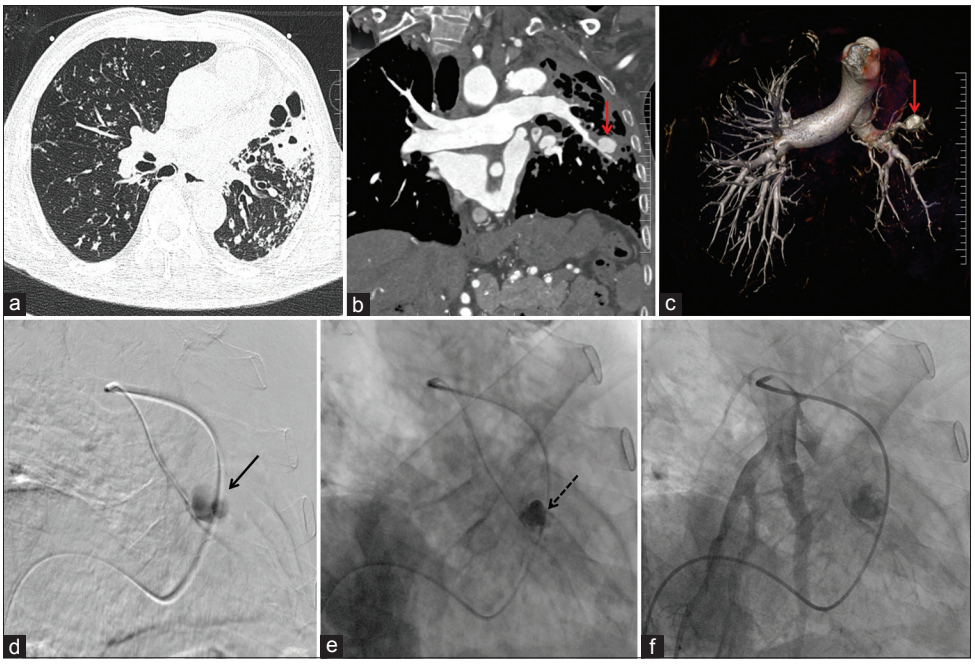


**Figure 2:** Axial section of high-resolution computed tomography chest (a) in lung window showing subsegmental areas of cavitatory consolidation in the left lingula and left lower lobe with multiple discrete and confluent centrilobular nodules in visualized bilateral lung fields, many of them showing tree in bud appearance. Coronal reformatted section of contrast enhanced computed tomography chest (b) showing 15 mm × 13 mm sized pulmonary artery pseudoaneurysm noted arising from anteromedial basal segmental branch of the left pulmonary artery (red arrow). Three-dimensional volume-rendered image of computed tomography pulmonary angiography (c) showing pulmonary pseudoaneurysm (red arrow). Right anterior oblique projection of catheter pulmonary angiography (d) selective injection of contrast after cannulation of segmental branch showing opacification of pseudoaneurysm. Right anterior projection fluoroscopic image (e) showing glue injection in pseudoaneurysm (dashed black arrow). Postglue embolization anteroposterior projection of catheter angiography (f) showing glue cast in pseudoaneurysm with normal opacification of rest of the left descending pulmonary artery

size, number, shape, and origin of pulmonary artery aneurysm/pseudoaneurysm. In addition, the presence of dilated, tortuous, and hypertrophied bronchial arteries is easily identified. Nonbronchial systemic arteries or ectopic bronchial arteries if any are also identified.

The treatment options for pulmonary artery pseudoaneurysms include open surgical and percutaneous techniques (CT-guided percutaneous embolization and endovascular techniques). No randomized control trial is there to suggest efficacy of surgical excision or endovascular treatment over each other.[4]

Surgical management, especially in emergency setting has high morbidity and mortality with prolonged patient recovery, hospital stay, and decreasing pulmonary reserve.[4,6] In addition, endovascular techniques obviate the need of general anesthesia and its associated complications.

Endovascular techniques provide with the advantages of being minimally invasive, safe, and effective with reduced morbidity and mortality. When percutaneous approach is used, there is always a question whether to embolize pulmonary or systemic arteries in patients with massive hemoptysis. Remy *et al*. suggested that when a pulmonary arterial source of bleeding is suggested, the first thing to embolize is the pulmonary artery lesion.[1] Similar results were found in study by Sbano *et al*.[7] Shin *et al*. suggested combined embolization of pulmonary and bronchial arteries in cases with infective etiology, suspicion of bronchopulmonary arterial shunt and too small pulmonary artery pseudoaneurysms.[8]

Various endovascular catheter-directed treatment techniques have been described and include coil embolization using coils or microcoils with or without bare stents (caging technique), use of microvascular plug, and embolization using liquid embolic agents. Complications of endovascular embolization are less frequent and include contrast-induced nephropathy, arterial dissection, and pulmonary infarct.

Coil embolization of pulmonary pseudoaneurysms is previously described in few case reports and miniseries.[9] They offer the potential for selective embolization of the sac while preserving flow through the affected pulmonary artery segment, however, are associated with risk of distal coil migration in case of aneurysms with a wide neck. In these cases, use of bare stents (caging technique) may help, however, increases the procedure costs. In addition, placing coils within the aneurysmal sac increases the risk of rupture.[4] Another technique used is coil embolization of pulmonary

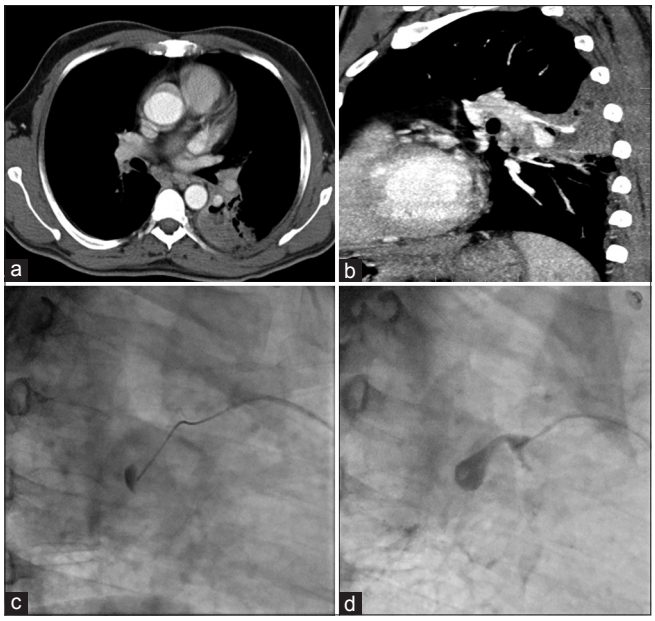


**Figure 3:** Axial and sagittal reformatted sections of contrast-enhanced computed tomography chest (a and b) in mediastinal windows showing subsegmental area of consolidation in superior segment of the left lower lobe with two pulmonary artery pseudoaneurysms noted arising from superior segmental branch of the left pulmonary artery, larger measuring 11 mm × 9 mm. Oblique projection of catheter pulmonary angiography (c) selective injection of contrast after cannulation of segmental branch showing opacification of pseudoaneurysm. Oblique projection fluoroscopic image (d) showing glue injection in pseudoaneurysm

artery branch supplying the aneurysmal sac. In such cases, the presence of bronchopulmonary shunt may still allow perfusion; however, simultaneous bronchial artery embolization would encourage pseudoaneurysm thrombosis.[7]

Jagia *et al*. suggested the use of vascular plug (patent ductus arteriosus closure device) for the treatment of pulmonary pseudoaneurysm. Amplatzer vascular plug has advantages of single device occluding the feeding vessel, increased precision, and control while deployment and firm anchorage to vessel wall by outward radial force due to elasticity of nitinol. They are difficult to be used in case of peripheral pulmonary arteries in view of large profile of access and deploying devices.[4]

Liquid embolic agents used for the treatment of pulmonary artery pseudoaneurysms include glue (N-butyl cyanoacrylate) and thrombin. Glue provides with advantages of being permanent, and highly effective; however, there have been only few case reports describing its use for treatments of PAP. Keeling *et al*. have reported that use of N-butyl cyanoacrylate is advantageous for PAP embolization because less time is needed and the risk for potential injury as compared to coil embolization is lower.[3] Endovascular coil embolization needs a stable microcatheter tip position in the aneurysmal sac which may be a problem, especially in dynamically pulsating pulmonary artery circulation having distorted anatomy due to underlying fibrocavitatory lung changes in clinical setting of Rasmussen's aneurysm; however, use of cyanoacrylate glue may be technically more feasible in such cases of tricky microcatheter position. There are risks of nontargeted embolization (proximally, distally, or into bronchus), pulmonary infarction or catheter adhesion.[4] A detailed evaluation of vascular anatomy and flow pattern obtained using test injections may reduce these risks. Minor reactions in the form of fever, nausea and vomiting (postembolization syndrome) may occur and respond to conservative management.[10]

Percutaneous CT-guided or US-guided treatment using thrombin cyanoacrylate glue injection can be done in cases refractory to endovascular technique of management.[11]

Our results demonstrate that glue (N-butyl cyanoacrylate) can be satisfactorily used for emergent management of pulmonary artery pseudoaneurysms presenting as massive hemoptysis. Endovascular treatment, especially with glue embolization, is less commonly practiced till date. In our series, the procedure has 100% technical success rate and absence of major complications. Transcatheter embolization currently is preferred for immediate therapy and surgical resection reserved for failed embolization or repeated recurrence. Choice of endovascular embolization material should be dictated by comfort and experience of operator and anatomical factors.

## CONCLUSION

Massive hemoptysis is a clinical emergency and needs prompt management. Pulmonary artery pseudoaneurysms are a rare cause of massive hemoptysis. Endovascular glue embolization is a technically feasible, effective, and minimally invasive treatment option.

The study has a limited sample size, and studies with larger sample size are needed to evaluate the efficacy and safety of endovascular glue embolization of PAP. In addition, the mean follow-up was 11.7 months in this study which is relatively short.

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Nil.

### Conflicts of interest
There are no conflicts of interest.

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