



Case report

Cement pulmonary embolism after percutaneous vertebroplasty in a patient with cushing's syndrome: A case report

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ABSTRACT

Background: Vertebroplasty is a procedure most commonly used for vertebral compression fractures. Although it is a relatively safe procedure, complications have been reported. Cement embolism is seen in 2.1%–26% of patients after percutaneous vertebroplasty.

Case presentation: a 38-year-old male who was diagnosed with cushing's syndrome, underwent percutaneous vertebroplasty for his thoracic osteoporotic compression fractures. 24-hours following vertebroplasty, he presented to emergency department with acute-onset dyspnea and chest pain. Chest radiography showed an opaque linear lesion in left pulmonary artery which was suggestive of cement embolism. Pulmonary spiral CT-scan further confirmed the diagnosis. The patient's symptoms improved over time, and warfarin was started with close cardiopulmonary assessments for indicators of cement embolus removal.

Conclusion: in patients with pulmonary cement embolism, conservative treatment may be recommended rather than a surgical removal except when the obstruction is extensive enough to cause hemodynamic changes. Given that all the related studies have suggested that pulmonary thromboembolism can occur as a complication due to bone cement leakage, discovering new cement alternatives and/or injection devices, seems beneficial.

1. Background

Vertebroplasty is a minimally invasive procedure most commonly used for vertebral compression fractures which was first introduced by Galibert et al., in 1987 [1]. In this procedure, polymethylmethacrylate (PMMA) is injected directly into the vertebral body through its pedicle, to restore the height partially, stabilize bony trabeculae, and alleviate pain. Due to its minimal invasion and immediate pain relief, percutaneous vertebroplasty gained popularity for the treatment of painful tumor infiltration disease such as multiple myeloma [2], and metastatic carcinoma [3–5], and for patients who have refractory pain due to osteoporotic thoracolumbar compression fractures [6–8] Although it is a relatively safe procedure, complications have been reported [9,10]. Acrylic cement of polymethylmethacrylate injected into the vertebral body can leak into the paravertebral venous system and reach the pulmonary artery via the azygos vein leading to a cement pulmonary embolism [11–15]. Pulmonary embolism of cement is seen in 4.6% of patients after percutaneous vertebroplasty. It can be asymptomatic and is directly related to the frequency of paravertebral venous leak, but not to the number of vertebral bodies treated [16]. Here, we report a case of

cement pulmonary embolism following vertebroplasty for thoracic compression fracture.

2. Case report

This is a 38-year-old smoker male who is a truck driver. He visited his family physician in July 2017, because of unintentional weight gain and a debilitating back pain. In physical examination he had a buffalo hump and central obesity, thus he was prescribed symptomatic treatment for his back pain and referred to an endocrinologist to evaluate for cushing's syndrome. His laboratory studies in following month showed a significantly high level of 24-h urinary free cortisol which was repeated 3 times and a plasma ACTH of 82pg/ml, which was suggestive of an ACTH-dependent cushing's syndrome. The urinary free cortisol after low and high-dose dexamethasone suppression test reported to be 546 and 764 mcg/24h respectively, which means resistance to dexamethasone and a negative test result. A magnetic resonance image (MRI) of pituitary following gadolinium administration was done which showed no abnormality. Because of the discordance between pituitary MRI, plasma ACTH level, and high-dose dexamethasone suppression

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test results, inferior petrosal sinus sampling (IPSS) was done by interventional radiologist, which showed a petrosal/peripheral ACTH ratio of less than 2. An ectopic ACTH syndrome was suggested which could not be localized with chest and abdominal CT scan. Ketoconazole was administered to control the cortisol excess, while planning for a bilateral adrenalectomy. The patient was also evaluated for his refractory back pain. MRI revealed diffuse osteopenic signal changes in lumbar vertebrae and multiple sites of compression fracture in all thoracolumbar vertebral bodies. Bone densitometry showed osteoporosis most severe at spine (mean Z-score and T-score < -2.9). As the patient was symptomatic, the decision has been made to proceed with vertebroplasty. High viscosity cement was injected into T7 to T12 vertebral bodies under fluoroscopic guidance in February 2018. The total volume of injected cement was 4 cc in each level. The patient tolerated the procedure and was discharged uneventfully. 24-hours following his vertebroplasty, he presented to our emergency department with a history of sudden-onset dyspnea and chest pain. Vital signs were within normal limits except tachycardia. He had no hypoxia, fever, chills, cough, and hemoptysis. The ECG was normal, except sinus tachycardia and cardiac troponins were negative. Echocardiography revealed no regional wall motion abnormalities with a 50% ejection fraction, a tricuspid valve regurgitation, and mildly increased systolic pulmonary artery pressure (35 mmHg). Chest radiography showed an opaque linear lesion in the left pulmonary artery (Fig. 1), which raised the suspicion of bone cement pulmonary embolism. Parenteral anticoagulation was started, and patient underwent pulmonary spiral CT-scan which revealed artifact-like hyperdense area in main pulmonary artery and left pulmonary artery suggestive of cement embolism (Figs. 2 and 3). During the hospitalization, patient's symptoms resolved, and warfarin was started. Cardiovascular surgery consultants recommended medical rather than surgical treatment with close cardiopulmonary monitoring for any signs and symptoms suggestive of worsening embolism. The patient was asymptomatic when he was discharged. Serial cardiac and pulmonary assessments will be carried out looking for increased pulmonary artery pressure as an indicator for the removal of the cement embolus.

3. Discussion

We present a case of 38-year-old man who underwent a T7 to T12 vertebroplasty because of osteoporotic compression fractures, and subsequently had a pulmonary cement embolization to his pulmonary arterial circulation, which was treated non-operatively with anticoagulation.

Operative treatment of vertebral compression fractures has included



Fig. 1. Chest X-ray showing linear opaque lesion in left pulmonary artery.



Fig. 2. Hyperdense lesion on left pulmonary artery.



Fig. 3. Hyperdense lesion on main pulmonary artery and left pulmonary artery.

percutaneous vertebroplasty for the past 30 years. Introduced by Galibert et al. [1] in 1987, this procedure gained popularity steadily and is used as an immediate pain relief method, in osteoporotic compression fractures [6–8] and for treatment of tumor infiltration disease such as metastatic carcinoma [3–5], and multiple myeloma [2]. Efficacy of vertebroplasty in alleviating pain, is not without controversy according to Buchbinder et al. [17] and Kallmes et al. [18] studies, which showed no improvement in pain and pain-related disability in osteoporotic spinal fractures.

Bone cement leakage is of particular concern. Cement leakage into the spinal canal can lead to canal stenosis and cord compression [19,20], and cement leakage into the intervertebral foramina can cause nerve root compression [21]. Additionally, cement leakage into the perivertebral system and inferior vena cava (IVC) can drift toward the right heart and pulmonary arterial system with catastrophic results such as cardiopulmonary arrest [33,34], acute kidney injury [22], paradoxical embolism through a patent foramen ovale [23], and death [10,24,36]. Arterial embolization to the aorta and anterior spinal artery has also been described [25,26]. The risk of cement pulmonary embolism first reported by Padovani et al. [27] exists with both vertebroplasty and kyphoplasty, but the exact rate is uncertain because the patients are not routinely screened for cement embolism [28]. The incidences of pulmonary cement embolism after vertebroplasty ranges from 2.1% to 26%, with much of this variation resulting from which imaging technique is used and whether the study is prospective or retrospective [16,29–32]. Clinical features of cardiopulmonary side effects of cement leak in percutaneous vertebroplasty and kyphoplasty include precordial chest pain and tightness [33–36], dyspnea [35–38], cyanosis, palpitation [34], acute respiratory distress syndrome (ARDS)

Table 1
Published case reports of pulmonary/cardiac cement embolism (1999–2017).

Outcome	Treatment	Clinical manifestation	Indication	Gender	Age (years)	Author/Publication date
Uneventful recovery	Anticoagulant + Supportive oxygen	Chest pain Hemoptysis hypoxia	Chronic osteoporotic pain	F	41	Padovani et al. (1999) [27]
Uneventful recovery	Anticoagulant unclear	Sudden onset dyspnea	Osteoporotic fracture unclear	unclear	unclear	Perrin et al. (1999) [61]
Recovered from respiratory and cardiac failure	Anticoagulant + Embolectomy	2 asymptomatic patients and 1 symptomatic patient Respiratory distress, atrial fibrillation, hypoxia	Pathologic fracture (osteogenesis imperfecta)	M	55	Amar et al. (2001) [62] Tozzi et al. (2002) [39]
Uneventful recovery	Supportive oxygen + Anticoagulant	Mild dyspnea and chest discomfort	Compression fracture	M	60	Jang et al. (2002) [50]
Died	CPR for 60 minutes	dyspnea and chest discomfort	Compression fracture	M	57	
Uneventful recovery	Anticoagulant + Embolectomy (interventional catheter procedure + open heart operation)	Asymptomatic	Compression fracture	F	60	
discharged	No treatment	Sudden onset bradycardia, shock, hypercapnia	Osteoporotic fracture	F	52	Chen et al. (2002) [24]
	Anticoagulation	Mild dyspnea Large mass on X-ray	Compression fracture	F	52	Francis et al. (2003) [38]
	Anticoagulation	No symptom	Multiple compression fractures	M	67	Bernhard et al. (2003) [44]
Respiratory symptoms improved	Ventilation + 6 mg hydrochloride Ephedrine	Repiratory and cardiac distress	Osteoporotic fracture	F	62	Torres Machi et al. (2003) [63]
Died	Mask ventilation, positive pressure ventilation, repeated intravenous boluses of noradrenaline and adrenaline	Severe chest pain, Restless, tachypnea, tachycardia, hypertension, oxygen desaturation, loss of consciousness, pulseless electrical activity	Osteoporotic fracture	F	62	Charvet et al. (2004) [64]
Died	tracheal intubation and mechanical ventilation, intravenous anticoagulation, pulmonary embolectomy	Arthralgia, myalgia, fever, ARDS	Osteoporotic fracture	F	68	Stricker et al. (2004) [45]
Discharged	Anticoagulation	Asymptomatic	Osteoporotic fracture	F	80	Yoo et al. (2004) [40]
Discharged	Open heart surgery for hemopericardium and cement removal	Chest pain, hemopericardium, cardiac perforation	Osteoporotic fracture	F	80	Pleser et al. (2004)
Respiratory symptoms improved	Right atriotomy and inferior vena cavotomy	Palpable mass on the subareolar of the left chest wall	Compression fracture	M	72	Kim et al. (2005) [33]
died	Anticoagulation	Sudden onset dyspnea	Osteoporotic fracture	F	78	Seo et al. (2005) [65] Pott et al. (2005) [66]
Discharged	Anticoagulation	Shock, hypoxia, cardiac arrest	Pain	F	81	Monticelli et al. (2005) [12]
Discharged	Anticoagulation	Asymptomatic	Osteoporotic fracture	F	50	Baummann et al. (2006) [67]
Discharged	Anticoagulation	Hypotension Arrhythmia hypoxapnia	Collapsed vertebra, pain	F	63	Freitag et al. (2006) [49]
Discharged	No treatment	Respiratory distress	Osteoporotic fracture Due to multiple myeloma	F	65	MacTaggart et al. (2006)
Died	Anticoagulant	Dyspnea, chest pain	Bone metastasis	F	68	Barragan-Campos et al. (2006) [10]
Uneventful recovery	Anticoagulant	Asymptomatic	Osteoporotic fracture	F	45	Abdul-Jalil et al. (2007)
Discharged	No treatment	Dyspnea, chest pain	Osteoporotic fracture	F	64	[48]
Uneventful recovery	No treatment	Asymptomatic	Lumbar hemangioma	M	61	Bonardel et al. (2007) [68]
Uneventful recovery	No treatment	Dyspnea Cough Chest pain	Osteoporotic fracture	F	85	Liliang et al. (2007) [51]
Uneventful recovery	Anticoagulant	Mild dyspnea	Osteoporotic fracture	F	55	Lim et al. (2007) [37]
Uneventful recovery	Pericardial collection aspiration	Chest pain Dyspnea Pericardial effusion	Compression fracture	F	59	Lim et al. (2008) [35]
Uneventful recovery	Right ventricular wall repair Tricuspid annuloplasty	Chest pain Chest tightness Hemopericardium Severe TR Cardiac tamponade	Compression fracture	F	65	Son et al. (2008) [56]
Discharged	Right cardiac catheterization Failed cement removal	Asymptomatic	Osteoporotic fracture	F	68	Cadeddu et al. (2009) [47]
Uneventful recovery	Endovascular cement removal	Chest pain palpitation	Bone metastasis (pain)	F	51	Braiteh et al. (2009) [34]
Uneventful recovery	Anticoagulant	Progressive dyspnea Tamponade	Osteoporotic fracture	F	64	Caynak et al. (2009) [36]
Uneventful recovery	Anticoagulant	Asymptomatic	Severe scoliosis and pain	M	76	Akinola et al. (2010) [69]

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Table 1 (continued)

Outcome	Treatment	Clinical manifestation	Indication	Gender	Age (years)	Author/Publication date
Discharged	Conservative management	Dyspnea Cough	Osteoporotic fracture pain	F	79	Radcliff et al. (2010) [70]
Reported asymptomatic and clinically silent patients with PCE in 26% of patients treated with PVP						Vennans et al. (2010) [31]
Reported 23 cases of PCE after PVP in 244 patients whom 1 patient was symptomatic from PCE						Luetmer et al. (2011) [41]
discharged	Anticoagulant oxygen	Hypoxia	Osteoporotic fracture	F	78	Abd El-Rahman et al. (2012) [71]
Died	Percutaneous retrieval of large cement fragment Mechanical ventilation	Multiple pulmonary embolies seen in fluoroscopy ARDS Pneumonia	Fracture due to bone metastasis	M	74	Alcibar et al. (2012) [72]
Uneventful recovery	Anticoagulant	Pleuritic chest pain Dyspnea	Compression fracture due to bone metastasis	F	37	Chick et al. (2012) [73]
Uneventful recovery	Lidocaine Amiodarone Magnesium sulfate Surgical cement removal	Non-sustained ventricular tachycardia Right ventricular failure Foreign body in right ventricle	Fracture due to bone metastasis	F	65	Cohen et al. (2012) [74]
Uneventful recovery	IVC filter Thrombectomy Urokinase Anticoagulant Balloon angioplasty	Leg swelling Thrombosis extending from IVC to right common iliac vein Subsegmental pulmonary embolism	Osteoporotic fracture	M	69	Kim et al. (2012) [75]
Uneventful recovery	Anticoagulant	Chest pain Tachypnea tachycardia	Osteoporotic fracture	F	63	Liu et al. (2012) [76]
Discharged	No treatment	Asymptomatic	Osteoporotic fracture	F	83	Matouk et al. (2012) [77]
Not clear	Not clear	Asymptomatic	Osteoporotic fracture	F	50	Mishriki et al. (2012) [78]
Uneventful recovery	No treatment	Dyspnea	Osteoporotic fracture	F	82	Bopparaju et al. (2013) [79]
Died	Anticoagulant Failure of complete cement removal	Sudden dyspnea Tricuspid regurgitation	Fracture due to multiple myeloma compression fracture	F	62	Chou et al. (2013) [80]
Not clear	Not clear	Asymptomatic	compression fracture	M	69	Garcia-Fontan et al. (2013) [81]
Uneventful recovery	Anticoagulant Antibiotic Short-term corticosteroids	Asymptomatic	Osteoporotic fracture	M	70	Geraci et al. (2013) [82]
Discharged	Open-heart surgery Cement removal	Dyspnea Chest pain Right Ventricle perforation	Fracture due to multiple myeloma	F	58	Gosev et al. (2013) [83]
discharged	No treatment	Dizziness Generalized weakness	Compression fracture	M	58	Lee et al. (2013) [84]
Uneventful recovery	Cardiopulmonary bypass surgery Cement removal from left lower lobe artery	Dyspnea Chest pain	Osteoporotic fracture	F	68	Llanos et al. (2013) [85]
Uneventful recovery	Surgical removal	Pericardial effusion Fever leukocytosis Chest pain asymptomatic	Osteoporotic fracture	F	86	Moon et al. (2013) [86]
discharged	No treatment		Osteoporotic fracture pain	M	74	Sifuentes et al. (2013) [87]
Not clear	Anticoagulant	Dry cough	Osteoporotic fracture	F	76	Yu et al. (2013) [88]
Uneventful recovery	Surgical removal of cement	Hypotension ARDS	Traumatic compression fracture	F	71	Arnaiz-Garcia et al. (2014) [89]
Uneventful recovery	Antiviral No treatment for PCE	Acute respiratory failure CMV pneumonia	Fracture due to bone metastasis	M	49	Chebib et al. (2014) [90]
discharged	No treatment	asymptomatic	fracture due to bone metastasis	F	39	Chen et al. (2014) [91]
Transferred to other hospital	Anticoagulant Inotrops	Intermittent dyspnea hypotension	Osteoporotic fracture	F	56	Huh et al. (2014) [92]
discharged	Open-heart surgery	Progressive dyspnea hemothorax	Chronic back pain	F	68	Kim et al. (2014) [93]
Uneventful recovery	No treatment	Syncope	Fracture due to multiple myeloma	F	52	Pannirselvam et al. (2014) [94]
Discharged	Cardiopulmonary bypass Embolectomy Pulmonary wedge resection	Chest pain	Traumatic fracture Pain	M	29	Rothermich et al. (2014) [95]
Discharged	No treatment	Dull chest pain	Pathologic compression fracture	M	60	Stevens et al. (2014) [96]
Uneventful recovery	Antibiotic Anticoagulant	Dyspnea	Compression fracture due to exogenous cushioning	M	48	Toru et al. (2014) [97]
Uneventful recovery	Percutaneous retrieval of cement	Asymptomatic	Osteoporotic fracture	F	55	Zhao et al. (2014) [98]

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Table 1 (continued)

Outcome	Treatment	Clinical manifestation	Indication	Gender	Age (years)	Author/Publication date
Discharged	No treatment	Asymptomatic	Painful fracture	M	72	Guiguins et al. (2015) [99]
Discharged	No treatment	Dyspnea responding to nitroglycerine	Compression fracture pain	F	70	Nooh et al. (2015) [100]
Discharged	Antiplatelet	Dyspnea Chest pain	Not clear	F	69	Poll et al. (2015) [101]
Discharged	Open-heart surgery	Sudden onset chest pain	Chronic back pain	M	65	Schuerer et al. (2015) [102]
Uneventful recovery	Cardiopulmonary bypass			F	63	Shen et al. (2015) [103]
Discharged	Right atriotomy			M	70	Shroff et al. (2015) [104]
Not clear	Anticoagulant	Dyspnea	Traumatic fracture	F	54	Awwad et al. (2016) [105]
Discharged	Not clear	Palpitation Chest pain	Osteoporotic fracture	F	51	Chai et al. (2016) [106]
Discharged	Anticoagulant	Asymptomatic	Fracture due to bone metastasis	F	51	Chai et al. (2016) [106]
Uneventful recovery	Open-heart surgery	Dyspnea Hemopericardium	Osteoporotic fracture	M	28	Diab et al. (2016) [107]
Discharge	Open-heart surgery	Dyspnea	Traumatic fracture	M	64	Focardi et al. (2016) [108]
Discharge	No treatment	Asymptomatic	Osteoporotic fracture	F	58	Gabe et al. (2016) [109]
Discharge	No treatment	Asymptomatic	Fracture due to multiple myeloma	F	58	Gorospe et al. (2016) [110]
Discharge	No treatment	Asymptomatic	Not clear	M	32	Memarpour et al. (2016) [111]
Uneventful recovery	Endoscopic Robot-assisted open heart surgery	Chest pain Tachycardia	Osteoporotic fracture pain	F	72	Molloy et al. (2016) [112]
Uneventful recovery	Open-heart surgery	Hypotension Pericarditis Atrial fibrillation	Compression fracture	M	49	Park et al. (2016) [113]
Not clear	Not clear	Chest pain Right ventricular penetration	Osteoporotic fracture	F	77	Botia Gonzalez et al. (2017) [114]
discharged	Anticoagulant	Dyspnea	Traumatic compression fracture	M	59	Chang et al. (2017) [115]
Not clear	Not clear	Asymptomatic	Traumatic compression fracture	M	65	Gianculli et al. (2017) [116]
Uneventful recovery	Anticoagulant	Palpitation	Osteoporotic fracture	F	57	Hatzantonis et al. (2017) [117]
Uneventful recovery	Anticoagulant	Chest pain Pleural effusion	Bone neuro-ectodermal tumor	F	15	Ramanathan et al. (2017) [115]
Uneventful recovery	Steroids Anticoagulant	Fever Respiratory distress hemoptysis	Fracture of femur	F	96	Talec et al. (2017) [118]
Uneventful recovery	Anticoagulant	Hypoxemia	Not clear	M	57	Wu et al. (2017) [13]
Uneventful recovery	Anticoagulant Surgical removal	Dyspnea Chest pain				

M = male, F = female, PCE = pulmonary cement embolism, ARDS = acute respiratory distress syndrome, PVP = percutaneous vertebroplasty.

[39,40], and cardiac arrest [12], although some patients with pulmonary cement embolism are asymptomatic [41–44]. The symptoms of cement embolism occurs more commonly days to months after, rather than during the procedure [12,24,39,45]. The cement used in vertebroplasty is of such high density compared to lung field that the visualization of cement emboli on CXR is quite striking, but multiple dense opacities with a branching shape which are scattered randomly or diffusely throughout the lungs are more common [16,29,44]. In our patient, CXR showed an opaque linear lesion in the left pulmonary artery without significant scattered lesions in the lungs. Echocardiography is a safe and non-invasive modality to evaluate hemodynamic status and to reveal the probable echogenic material in the cardiac chambers [46,47]. Chest CT scan accurately shows the locations, the lengths, and the number of cement emboli [35].

Abdul-Jalil et al. proposed that PMMA has a prothrombotic property and can cause endothelial injury, which can result in additional thrombosis [48]. The formation of PMMA toxins can cause direct cellular injury by increasing membrane permeability through releasing inflammatory mediators, and superoxide production. Pulmonary cement embolism finally shares similar pathophysiological similarities with pulmonary embolisms [40].

The cornerstone of treatment of pulmonary cement embolism is close cardiopulmonary monitoring and anticoagulation [27,49–53] but there are some reports of cement embolism requiring surgical removal (including cardiopulmonary bypass and arteriotomy) [33,35–39,54–56]. Choe et al. proposed that asymptomatic pulmonary cement emboli should not alter medical treatment [16]. In Venman's study, all 11 patients with venous PMMA migration remained asymptomatic during 1-year follow up [31]. Krueger et al. proposed a management algorithm that includes conservative approach for peripheral asymptomatic cases, anticoagulation for the symptomatic peripheral and asymptomatic central emboli, and surgical treatment for symptomatic central embolism only [57]. We selected anticoagulation and close monitoring for our patient regarding the published case reports of cement embolism which is summarized in Table 1. Because of non-degradable and toxic properties of PMMA, attempts have been made to explore alternative materials that are more suitable for vertebroplasty and kyphoplasty [58–60].

4. Conclusion

In patients with pulmonary cement embolism, conservative treatment may be recommended rather than a surgical removal except when the obstruction is extensive enough to cause hemodynamic changes. Given that all the related studies have suggested that pulmonary thromboembolism can occur as a complication due to bone cement leakage, discovering new cement alternatives and/or injection devices, seems beneficial.

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Availability of data and materials

All data and materials described in the manuscript will be freely available to any scientist wishing to use them for non-commercial purposes.

Authors' contribution

Authors contributed equally to this paper.

Competing interests

The authors declare that they have no competing interests.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by the Editor of this journal.

Ethics approval and consent to participate

Not applicable.

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Appendix A. Supplementary data

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