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Fatal hemoptysis from coil migration after endovascular coil embolization of a pseudoaneurysm years after anterior thoracolumbar scoliosis correction despite the acceptable original position of the screw: An autopsy case report

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ABSTRACT

Introduction: Fatal hemoptysis due to pseudoaneurysm development of the aorta following spinal screw instrumentation to correct scoliosis is a very rare clinical presentation.

Case presentation: We present an autopsy case of a 75-year-old German woman with a rare clinical presentation due to delayed aortic pseudoaneurysm that developed over years by compression of a screw head that was appropriately positioned. This aneurysm occurred years after scoliosis correction using anterior spinal instrumentation and lateral vertebral body screws. She was admitted to the hospital with abdominal and back pain. Her computed tomography (CT) and magnetic resonance imaging showed a hematoma of the paravertebral muscle at level Th12. Subsequently, diagnostic angiography was performed, which showed a paravertebral pseudoaneurysm with an aortic laceration approximately 3 cm in size. Interim emergency coil embolization was performed. Before a definitive endovascular stent could be placed the patient demised due to fatal hemoptysis. The autopsy revealed aortic wall injury, intra-alveolar, fresh pulmonary hemorrhage, a detachable coil at the site of the aortic rupture and migration of coils to the pulmonary parenchyma. Change of the diaphragm with fistula. The head of the internal fixator screw was seen in the hole through the aortic wall.

Conclusions: If direct contact between the aorta and a fixation screw following spinal surgery is identified in the postoperative course, such a rare complication should be considered and appropriate imaging (CT) on regular follow-up should be performed.

1. Introduction

Fatal hemoptysis due to rupture of a delayed aortic pseudoaneurysm after spinal screw instrumentation to correct scoliosis is a very rare clinical presentation. To correct thoracolumbar scoliosis through fusion there are two options: posterior and anterior instrumentation surgery. Anterior instrumentation surgery provides a better correction and shorter fusion level along with less postoperative pain and scaring [1]. Video-assisted thoracoscopic surgery could be an alternative method of treatment. Recently, posterior instrumentation surgery has become the superior option for anterior surgery due to compatible correction, shorter length of surgery, fewer risks for vascular injury and shorter overall hospital stays [2].

Anterior instrumentation is relatively dangerous because the thoracic aorta is at risk if screws penetrate the cortex of the vertebral body on the opposite side, which can affect pulmonary function after surgery. Screw malposition, dislocation, and loosening have been reported in the literature, however, the number of reports is limited [2–5]. Aortic injuries and delayed aortic pseudoaneurysms following spinal screw instrumentation have also been reported [1]. Due to the high mortality rate associated with open aortic aneurysm repair, endovascular stenting followed by a prolonged course of anti-coagulants while the stent undergoes epithelialization has become the treatment of choice. As an interim measure, specifically in aortic pseudoaneurysms, coil embolization may be performed before definitive stenting can be performed

Most of the published complications related to screws are case reports; case series and retrospective studies are limited [3]. Rare and fatal hemoptysis due to aortic aneurysm ruptures through the diaphragm and fused pleurae into the lung have been reported [6].

We present a case of an abdominal aortic pseudoaneurysm cause by

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the dislocation of a spinal screw head. The patient's clinical presentation was abdominal pain followed by pulmonary hemorrhage and hemoptysis leading to death. The surgical complication in the present case differs from those described in previous reports in that the aneurysm occurred because of the over the year's dislocation of the screw head. The unique clinical presentation and autopsy confirmed the pathology.

2. Case presentation

A 75-year-old German female died from heart failure complicated by respiratory failure caused by hemoptysis. The patient presented to the emergency department with refractory abdominal and back pain with a radiculopathy associated with the L3 region. The initial computed tomography (CT), magnetic resonance imaging, and abdomen sonography imaging demonstrated a hematoma of muscle in the paravertebral area at the T12 level. Diagnostic angiography was performed that showed a paravertebral pseudoaneurysm. An aortic laceration of approximately 3 cm was found in the wall of the aorta. Because of the poor cardiac and pulmonary condition of the patient, an endovascular procedure was performed. Embolization with platinum was done (spiral 25 mm × 50 cm coil (Axium 3 D, Fa. EV3, 24 coil with 25 mm to 13 mm with length 30 cm). On the second day, the patient experienced hemoptysis with pulmonary hemorrhage and re-embolization was emergently planned but the patient died before the second procedure.

The patient's history indicated back pain that occurred following the last operation, which progressively increased for one month duration. The patient's clinical history revealed that at the age of 68 years, she first presented to the university clinic in north Germany with severe thoracolumbar left convex scoliosis. An operative correction was indicated and combined anterior instrumentation with lateral vertebral body screws and posterior instrumentation with pedicular screws was performed on the T8 to L3 region to prevent the progression of the curve. The operation was uneventful, and the patient recovered without complications. At the age of 72 years the patient presented to another clinic with spinal canal stenosis at T8/9 and subsequently, decompression and stabilization was performed at this region. At the age of 74, the patient underwent decompression and stabilization of L2 to L4 due to spinal canal stenosis pseudoarthrosis. Dorsal stabilization was not sufficient and 5 months later, ventral stabilization at the level L2-4 level was carried out. Once the patient was stable, she was discharged to the rehabilitation center. (Fig. 1)

3. Autopsy findings

After written consent from the patient's family, the autopsy was performed 24 h following her death. Standard autopsy procedures showed severe thoracolumbar scoliosis that was previously corrected with internal fixation surgery. In the lumbar position, the head of the screw was directed toward the abdominal aorta and was causing aortic wall injury. The head of the screw completely damaged the dorsal wall of the aorta causing massive retroperitoneal bleeding along the length of the aorta through the diaphragmatic section of the aorta into the left thorax. A pseudoaneurysm, also known as an aneurysm spurium, occurred due to the added aortic pressure. This eventually led to increased pressure in the pulmonary parenchyma, intra-alveolar hemorrhage followed by extreme blood loss through the tracheobronchial tree. The bleeding was managed with coiling. The intra-aortic pressure was very high, which prevented the coiling device from maintaining its position; the pressure also led to aortic perforation and subsequent bleeding through the diaphragmatic section of the aorta into the lung. The device added to the lung parenchyma damage causing a massive bleed. The patient died from cardiovascular failure due to massive bleeding and anemia.

On histological examination, the hematoxylin and eosin (HE)-staining (10:1 magnification) showed an overview of lung bleeding after the aortic rupture. HE-staining, at 100:1 magnification showed intra-alveolar hemorrhaging while the macroscopic findings showed coiling of the aorta rupture, an alteration of the diaphragm and the lung, internal fixation of the spine, and aneurysm spurium after aorta rupture was observed in the hole through the aorta wall. Alteration caused by the head of the screw used for internal fixation of the spine is shown in Figs. 1–4.

4. Discussion

Aortic pseudoaneurysms are a rare entity; potentially fatal causes of pseudoaneurysms are trauma, infection, atherosclerosis, iatrogenicities or a late complication of a previous surgical procedure [7]. Hemoptysis

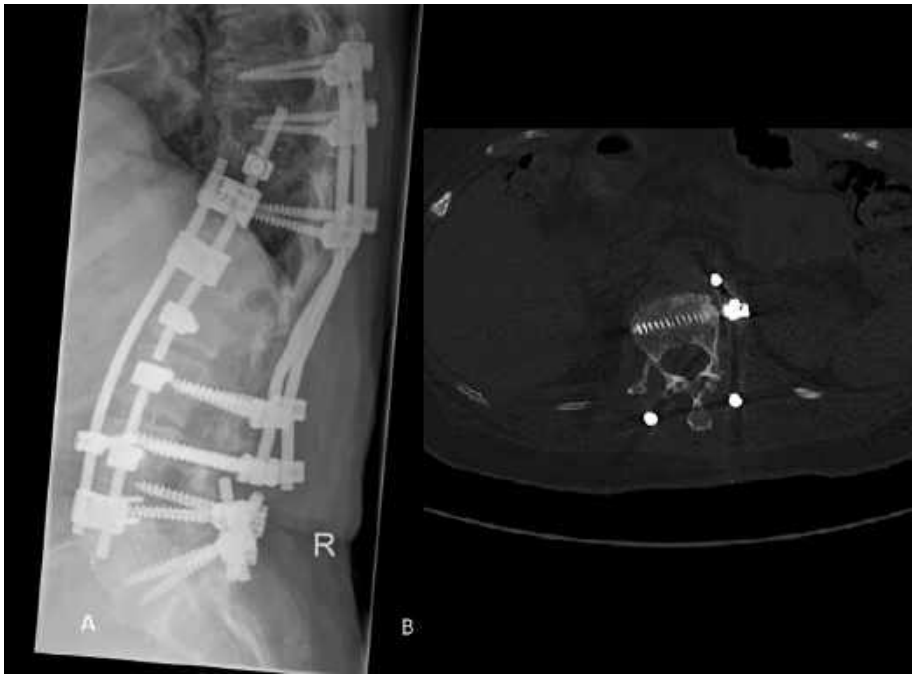

Fig. 1. (A) Anterior and posterior screw instrumentation to correct scoliosis. (B) Screw head responsible for the damaged the aorta.

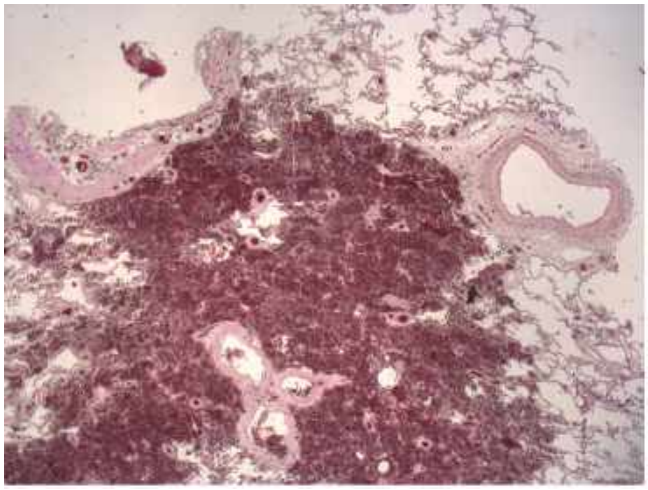

Fig. 2. Histopathology of the lung: Overview of lung bleeding after an aortic rupture. HE-staining, 10:1 magnification. Abbreviations: HE, hematoxylin and eosin.

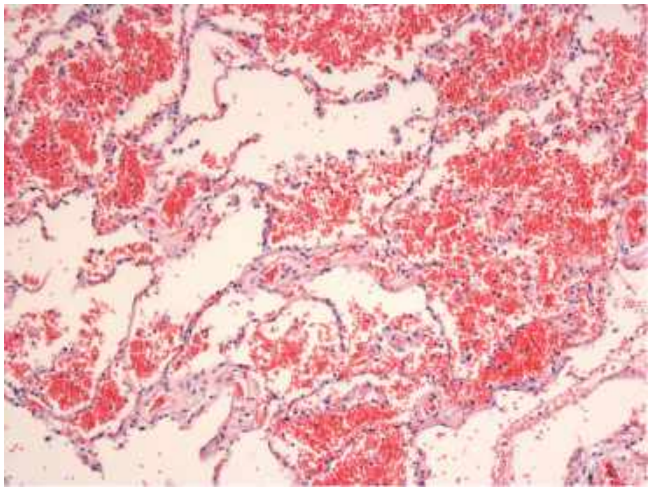

Fig. 3. Histopathology of the lung: intra-alveolar fresh bleeding after coiling of the aorta rupture. Alteration of the diaphragm and the lung, HE-staining, 100:1 magnification. Abbreviations: HE, hematoxylin and eosin.

Fig. 4. A: Aneurysm spurium after aortic rupture. Alteration caused by the head of the screw for internal fixation spine seen in the hole through the aorta wall. B: coils.

due to aortic aneurysms is an uncommon presentation; however, when it presents it is usually fatal. Very few cases of aortic aneurysms

Fig. 4. (*continued*)

presenting with hemoptysis have been published [8]. Rupture into the lung and erosion of the trachea with fistula formation is a very rare event, which has also been previously reported in the literature [9].

In our case, trauma due to direct contact between the screw head and the aorta was the cause of the pseudoaneurysm, although the screw was appropriately placed during the spine surgery, over a long period of time, direct contact with the aorta occurred, due to a phenomena called "screw pull out" and the aorta moving after scoliosis correction, thereby causing damage to the aorta.

Published data has shown a significant rate of rod breakage and proximal screw pullout associated with spine procedures; furthermore studies show that the aorta may move closer to the vertebral column after scoliosis correction [10,11].

There are several suggested surgical techniques to avoid this complication; facetectomies, mesh cages at proper disc spaces, using either single or double rods, hook claws and a claw for the top instrumented vertebra may be performed [12–15]

In addition, general erosion and laceration of the aorta by anterior spinal implants causing retroperitoneal hemorrhage, hematemesis, spontaneous pneumothorax, hemoptysis and the formation of a pseudoaneurysm have been reported [16,4,17].

Injury to the aorta may be due its anatomical position, which follows the spine even in severe scoliosis; the aorta adheres to the abnormal curves of the spine [17].

Maruyama et al. reported that the aorta could be located in the direction of screw passage between Th6 and Th9 in 83% of the patients reported in their study. The aorta is fairly mobile at the midthoracic level when a patient changes position [18]. Aortic contact to the screw was noted in 0.07% of the 8147 screws in eight studies; however, most of the information is reported as case reports [3]. A substantial difference in the position of the aorta relative to the spine in the prone and supine position seen at levels Th4 to Th8 is another anatomical challenge [19].

The available management of aortic pseudoaneurysms is direct surgical repair, percutaneous coil embolization, stent-graft placement, thrombin injection, and use of Amplatzer devices [7]. Endovascular aortic repair showed significantly lower mortality rates than that seen with conventional open repair [20]. Location of the pseudoaneurysm, the size of the neck, origin of the great vessels and the general health condition of the patients are factors that determine the choice in type of endovascular treatment [7]. In our case, the patient was in an emergent situation, and therefore a complex and risky surgical operation could not be performed.

Aortic Stent-Graft was not placed in because the patient demised

due to fatal hemoptysis in the second day before a definitive endovascular stent could be placed. The Patient was at increased risk for infection with bad general condition. Moreover, coiling was adopted as bridge treatment because of the emergent situation [21]

Published data support performing endovascular minimally invasive techniques as the first choice in selected patients and may be adopted as a bridged treatment in emergent situations [20].

Endoleak is a complication of endovascular treatment and can result in continued aneurysm growth, potential rupture [22] and therefore regular radiographic follow-up is necessary [16].

A case report similar to ours discussed a patient with Marfan syndrome, who also had a delayed aneurysm that occurred 20 years after surgery in the descending thoracic aorta region caused by a Zielke device (Ulrich, Germany). Treatment of this aneurysm was also done by an endovascular approach [23].

Our case was different in terms of the clinical presentation of hemoptysis, the occurrence of a muscular hematoma, thoracic fistula development and the appropriate original position of the screw. In addition, we were able to confirm the pathology and identify that the erosion of the aorta was caused by the screw head.

5. Conclusion

This case represents a rare but potentially fatal complication of anterior spinal instrumentation surgery with an unusual presentation despite the acceptable original positioning of the spinal screw. Aortic wall erosion from the screw head is a risk for fatal complications. When spinal screws or other instrumentation come in direct contact with the aorta over prolonged periods of time, rare complications should be considered and appropriate imaging (CT) should be performed on regular follow-up.

6. Consent for publication

“Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.”

CRediT authorship contribution statement

Evariste Gafumbegete: Conceptualization, Investigation. **Alaa Eldin Elsharkawy:** .

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.inat.2020.100775.

References

[1] J.B. Choi, J.O. Han, J.W. Jeong, False aneurysm of the thoracic aorta associated with an aorto-chest wall fistula after spinal instrumentation, J Trauma - Inj Infect Crit Care 50 (1) (2001) 140–143.
[2] Di Silvestre M, Parisini P, Lolli F, Bakaloudis G. Complications of thoracic pedicle screws in scoliosis treatment. Spine (Phila Pa 1976) 2007; 32(15):1655-61.
[3] Hicks JM, Singla A, Shen FH, Arlet V. Complications of Pedicle Screw Fixation in Scoliosis Surgery: A Systematic Review. Spine (Phila Pa 1976) 2010;35(11):E465-70.
[4] Endrisak, Spontaneous abdominal aortic rupture from erosion by a lumbar spine fixation device: a case report, Surgery 99 (5) (1986) 631–633.
[5] J.R. Stewart, K.H. Barth, G.M. Williams, Ruptured lumbar artery pseudoaneurysm: an unusual cause of retroperitoneal hemorrhage, Surgery 93 (4) (1983) 592–594.
[6] Liljenqvist UR, Halm HFH, Link TM. Pedicle screw instrumentation of the thoracic spine in idiopathic scoliosis. Spine (Phila Pa 1976) 1997;22(19):2239-45.
[7] J. Hussain, R. Strumpf, G. Wheatley, E. Diethrich, Percutaneous closure of aortic pseudoaneurysm by Amplatzer occluder device - Case series of six patients, Catheter Cardiovasc Interv 4 (2009) 521–529.
[8] G. Nair, S. Jindal, A. Chandra, S. Swami, P. Garg, et al., Haemoptysis - a rare presentation of aortic aneurysm, Lung India 25 (2008) 20–21.
[9] A. Fujii, R. Sawamoto, Y. Misumi, D. Hidaka, H. Koshizuka, J. Hiyama, et al., An autopsy case of massive hemoptysis due to a rupture of the thoracic aortic aneurysm into the left lung, Nihon Kokyuki Gakkai Zasshi 47 (2009) 57–60.
[10] D.J. Sucato, F. Kassab, M. Dempsey, Analysis of Screw Placement Relative to the Aorta and Spinal Canal Following Anterior Instrumentation for Thoracic Idiopathic Scoliosis, Spine (Phila Pa 2004 (5) (1976) 554–559.
[11] V. Bullmann, E.M. Fallenberg, N. Meier, R. Fischbach, T.L. Schulte, W.L. Heindel, et al., Anterior dual rod instrumentation in idiopathic thoracic scoliosis: A computed tomography analysis of screw placement relative to the aorta and the spinal canal, Spine (Phila Pa 2005 (30) (1976) 2078–2083.
[12] C.G. Hopf, P. Eysel, J. Dubousset, Operative Treatment of Scoliosis With Cotrel-Dubousset-Hopf Instrumentation: New Anterior Spinal Device, Spine 22 (6) (1997) 618.
[13] Kaneda K, Shono Y, Satoh S, Abumi K. Anterior correction of thoracic scoliosis with Kaneda anterior spinal system: A preliminary report. Spine (Phila Pa 1976) 1997; 22(12):1358-68.
[14] A. Gogus, U. Talu, S. Akman, C. Sar, A. Hamzaoglu, L. Eralp, Anterior instrumentation for adolescent idiopathic scoliosis, Int Orthop 25 (5) (2001) 317–321.
[15] V. Arlet, J. Shilt, J. Ouellet, The suprapedicle claw construct in anterior scoliosis surgery, Eur Spine J 14 (8) (2005) 789–794.
[16] Watanabe K, Yamazaki A, Hirano T, Izumi T, Sano A, Morita O, et al. Descending aortic injury by a thoracic pedicle screw during posterior reconstructive surgery: A case report. Spine (Phila Pa 1976) 2010;35(20):E1064-8..
[17] M.E. Minor, N.J. Morrissey, R. Peress, A. Carroccio, S. Ellozy, G. Agarwal, et al., Endovascular treatment of an iatrogenic thoracic aortic injury after spinal instrumentation: Case report, J Vasc Surg (2004).
[18] Maruyama T, Takeshita K, Nakamura K, Kitagawa T. Spatial relations between the vertebral body and the thoracic aorta in adolescent idiopathic scoliosis. Spine (Phila Pa 1976) 2004;29(18):2067-9.
[19] Huitema GC, Cornips EMJ, Castelijns MH, Van Ooij A, Van Santbrink H, Van Rhijn LW. The position of the aorta relative to the spine: Is it mobile or not? Spine (Phila Pa 1976) 2007;32(12):1259-64.
[20] G. Piffaretti, M. Galli, C. Lomazzi, M. Franchin, P. Castelli, G. Mariscalco, et al., Endograft repair for pseudoaneurysms and penetrating ulcers of the ascending aorta, J Thorac Cardiovasc Surg 6 (2016) 1606–1614.
[21] G. Kicska, H. Litt, Preprocedural planning for endovascular stent-graft placement, Semin Intervent Radiol (2009).
[22] J.I. Fann, S. Samuels, S. Slonim, T.A. Burdon, R.L. Dalman, Treatment of abdominal aortic anastomotic pseudoaneurysm with percutaneous coil embolization, J Vasc Surg 35 (4) (2002) 811–814.
[23] Been HD, Kerkhoffs GMMJ, Balm R. Endovascular graft for late iatrogenic vascular complication after anterior spinal instrumentation: A case report. Spine (Phila Pa 1976) 2006;31(22):E856-8.