

An aortic wall to vertebral body fistula presenting as a lytic lesion

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ABSTRACT

There have been multiple reported cases of aortic fistulas but few cases of aorta to vertebral body fistulas and no aortic wall to vertebral body fistulas have been reported. Here we present a case of a patient who is status post thoracic aortic aneurysm (TAA) repair and found to have a lytic vertebral body lesion. Biopsy of the mass revealed blood products without evidence of malignancy and further investigation revealed a fistulous tract between the aortic wall and the vertebral body causing a vertebral body hematoma.

CASE REPORT

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A 69-year-old male who was status post complex repair of a thoracic aortic aneurysm fifteen years prior presented with multiple medical issues. He underwent a Crawford type 2 repair (extending from the subclavian to the iliac bifurcation) of an aneurysm which extended from the proximal descending aorta to below the renal arteries. The patient underwent routine surveillance post repair and aneurysm size remained stable for many years. He had multiple other comorbid conditions such as hypertension (HTN), Type 2 Diabetes Mellitus (DM), and Chronic Obstructive Pulmonary Disease (COPD) which also led to multiple admissions and repeat imaging of the thorax and abdomen.

During hospital admission in 2014, CT chest/abdomen revealed gross stability in the size aneurysm with the ascending aorta measuring up to 4.7 cm and descending aortic aneurysm measuring up to 4.8 cm with evidence of ulceration,

pseudoaneurysm, and thrombus. However, there was also evidence of a lytic L1 vertebral body lesion with sclerotic borders which was not seen on prior study (Figure 1a). The presumed diagnosis was metastatic disease and workup was undertaken to assess for primary malignancy.

Workup which consisted of additional imaging, colonoscopy, and labs such as light chain and electrophoresis was negative for malignancy. CT guided biopsy of the lesion was then performed which revealed only blood products. He was then discharged and re-presented 2 months later with flank pain and low blood pressure. Imaging at this time revealed large right retroperitoneal fluid collection with asymmetry of the psoas muscle. He was diagnosed with a large retroperitoneal hematoma. CT showed overall stability in the size of the aneurysm and CTA abdomen did not reveal any extravasation of contrast from the lumen of the aorta (Figs. 5-7). MRI of the lumbar spine with contrast revealed a T1 hypointense, T2 hyperintense, non-enhancing lesion

occupying most of the vertebral body with linear areas of low signal (Figs. 1b-4). Also, an apparent connection between the wall of the aorta and the process in the L1 vertebral body was recognized. The signal intensity of the vertebral body matched that of the TAA mural thrombus.

It was at this time hypothesized that there was a connection between the aorta and the vertebral body which caused replacement of the vertebral body with blood products. Since there was no active extravasation of contrast outside of the lumen on contrast enhanced studies, it was hypothesized that the fistulous connection was between the wall of the aorta and the vertebral body.

Unfortunately, as treatment options were being discussed, the patient passed away secondary to pulmonary complications. Post-mortem examination did confirm the hypothesis of a fistula of the abdominal aortic wall to the vertebral body.

Transverse sectioning through the superior abdominal aorta, celiac artery and L1 vertebra revealed tan-red soft tissue outside of the graft at the posterior aspect. Through a fistula tract, this tissue extended through the posterior periaortic tissue into the anterior L1 vertebral body (Figs. 8 and 9). The vertebral body and superior aspects of the bilateral transverse processes were hemorrhagic with central irregular areas of softening. A small psoas muscle hemorrhage was also identified adjacent to the left lateral aspect of the L1 vertebra.

Given the radiographic and pathologic findings, the patient appears to have developed a peri-anastomotic aneurysm in the lower thoracic Carrell patch with subsequent endoleak, type Ia (through the proximal suture site), leading to peri-graft hematoma and thrombus formation. The presence of focal graft degradation and acute inflammation of the graft and peri-graft hematoma may suggest a superimposed graft infection. The fistula tract connecting the posterior abdominal aorta to the L1 vertebra contained foci of inflammation, which may suggest a focus of infection, as well; however, the inflammatory infiltrates within the bone were not extensive. Special stains used to demonstrate the presence of bacterial organisms were not performed because premortem cultures were not collected and the autopsy was performed two days following death, raising the likelihood of polymicrobial contamination associated with decomposition.

DISCUSSION

Etiology & Demographics:

Before we can discuss the pathophysiology of how a fistula can form between the wall of the aorta and a vertebral body, we must discuss aneurysms and their potential complications. A true aneurysm is defined as a segmental dilation of a blood vessel having a true lumen which is at least 50 percent larger than the expected diameter. It must involve all three layers of the aortic wall (1). There are isolated thoracic and abdominal aortic aneurysms, and there are also aneurysms which involve both the thoracic and aortic regions.

The aneurysms involving both regions are classified as Crawford type 1-4 based on their degree of extension (2). Our patient had a type 2 aneurysm which extended from above the intercostal space to the infrarenal region. The estimated prevalence of asymptomatic TAA's has been reported at 0.16 and 0.34% depending on the study (1, 3). It most commonly occurs around 60-70 years of age and is 2-3 times more common in males (4). The risk factors of TAAs include, but are not limited to: bicuspid aortic valve, connective tissue disorders (Marfans and Ehlers-Danlos), aortitis, and risk factors for atherosclerosis such as: hypertension, smoking and hypercholesterolemia (5). It is unclear what role these factors cause in leading to aneurysmal formation, but it is hypothesized that these processes lead to cystic medial degeneration and weakening of the wall which allows for aneurysmal dilation from intraluminal arterial pressure (5).

Possible complications of aneurysms include: expansion, rupture, dissection, and fistula formation. One of the complications which can happen post repair is the endoleak. Endoleaks have been typically described after endovascular repair, but there have been reported cases of endoleak status post open repair. An endoleak is defined as continued arterial perfusion of an aneurysmal sac status post repair. An endoleak can have a number of different causes but is usually caused by poor seal of the fixation sites at the end of the grafts. It can also be caused by back flow of blood from arterial collaterals which are covered by the graft. Type 1 leaks are from incomplete seal at the proximal or distal ends of the graft. (Type 1a is at the proximal edge of the stent and Type 1B is distal). Type 2 is retrograde blood flow from a collateral artery which is adjacent to the covered stent. This backward flow fills the aneurysmal sac. Endoleak Type 3 are at points of connection between different components of the graft. Type 4 is wall porosity which means the fabric of the graft has allowed blood to seep through and communicate with the aneurysm sac. Type 5 is evidence of endoleak but unknown location.

Another complication is fistula formation. Primary aortic fistulas occur in the absence of prior aortic aneurysm repair and secondary aortic fistulas occur after aortic reconstruction. The mechanism of secondary fistula formation is proposed to be a product of inflammation from pulsation of the graft against the bowel wall. Also, pressurization of the aorta aneurysm sac secondary to endo leak which could also be a cause. Fistulas to abdominal organs such as small bowel have been well reported and are statistically the most common type. Most of the numbers for aortic fistulas after repair pertain to the aortoenteric fistula, and most common (75%) type of aortoenteric fistulas involve the duodenum (6). There are also some reports of fistulous connections between the aorta and vertebral venous plexus. However, there are few reports of aorta to vertebral body fistulas and no reported cases of aortic wall to vertebral body fistulas. Our patient has pathology to support a fistulous tract between the aortic wall and the vertebral body. Neither the lumen of the aorta or the aneurysmal lumen was shown to be communicating with the fistulous tract. It is thought that the tract was formed between the arteries or veins of the aortic wall (arterial or venous vasovascularum) and the vertebral body.

Clinical & Imaging Findings:

There are three main clinical presentations of abdominal aortic aneurysms, which are as follows: asymptomatic, symptomatic non-ruptured, and symptomatic ruptured (7). Symptoms typically include abdominal pain, back pain, or leg pain (8). Clinical signs can be tachycardia, hypotension, and laboratory evidence of end organ damage. Diagnosis of asymptomatic aneurysm can be made with many imaging modalities such as: CT, ECHO or MRI. The preferred method of diagnosis is CTA or MRA. Symptomatic aneurysms almost always undergo imaging by CTA. Once an aneurysm is diagnosed, a Class 1 recommendation is to follow aneurysms to assess for stability. There are multiple imaging standards set forth by the 2010 ACA/AHA guidelines which aims to reduce variability in the measurements from one study to the next (1).

Treatment & Prognosis:

Non-surgical management of TAAs consists mainly on blood pressure control and smoking cessation if the patient is a current smoker. Endovascular open surgical repair is usually recommended for symptomatic non-ruptured and ruptured cases. It is also generally recommended for size greater than 5.5 cm or less if the patient has a connective tissue disorder leading to the aneurysm. Also, if an aneurysm grows more than 0.5 cm in 1 year (Class 1), it may need to be treated, although this is variable and needs to be evaluated on a case by case basis (9).

The management of aortic fistulas usually consists of stabilizing the patient with volume resuscitation if the hemorrhage is significant. Also depending on site on fistulous connection, antimicrobials may be given prior to surgery. Surgery, either open or endovascular, is carried out to repair the aortic defect leading to the fistula. If it is a secondary fistula, the initial graft will need to be removed. Repair will also need to be provided to the adjacent organ involved. In our case the patient would have likely undergone repair of the aneurysm and then undergone repair of vertebral body. However, since the tract was not communicating with the lumen it is unclear if the graft would have been removed. If the patient survives aneurysm repair, the main cause of long-term death is cardiovascular disease, as was the case with our patient who passed away secondary to cardiopulmonary complications which led to PEA arrest (1, 9).

Differential diagnosis:

Metastatic disease is on the top of the differential diagnosis list for a newly diagnosed lytic bone lesion. On MRI, the lesion was T1 hypointense and T2 hyperintense which can be seen in certain malignant processes. The most common lytic metastases overall are breast cancer and lung cancer. These processes can also present as multifocal lesions and the patient may experience other symptoms related to primary malignancy. Diagnosis usually consists of whole-body imaging with skeletal series, bone scan, or FDG-PET/CT scan. Most metastatic processes are FDG-avid on PET (10). Ultimately, searching for the primary malignancy and biopsy of the lesion leads to the final diagnosis.

A malignant atypical hemangioma is also in the differential diagnosis for a process that can cause lytic spinal

lesions. Hemangiomas are benign tumors of vascular endothelial cells but can rarely degenerate to malignant forms. They have a characteristic striated appearance on CT from destruction of some trabeculations and thickening of others. They are traditionally hyperintense on T1 and T2 weighted imaging; however, vertebral hemangiomas can have atypical radiologic features (13, 14). These tumors are usually very slow growing and can produce symptoms of cord compression, pathologic fracture, or shunt like symptoms if vascular flow is high enough (14).

Aneurysmal bone cysts are expansile vascular lesions. They are usually solitary and can grow rapidly. Most occur in adolescents and are most commonly found in long bones. However, 20-30 percent can be found in the spine. CT shows lytic lesion with sharply defined thin sclerotic border. MRI signal intensity is variable but usually high intensity on T1 and T2 weighted imaging. MRI may also show the characteristic fluid-fluid level of ABC's (15).

Multiple myeloma/ plasmacytoma is also on the differential list. Multiple myeloma is a malignancy which arises from the bone itself as a clonal expansion of plasma cells. It usually appears as multiple punched out, lytic lesions on CT or x-ray. PET/CT is another option for diagnosis as it may show increased uptake in punched out, lytic areas. However, myeloma may also be "cold" on PET imaging, meaning it does not light up. MRI can show a change in marrow signal from the lesions. Typically myeloma is hypointense to isointense of T1 weighted imaging and isointense to hyperintense on T2 (11). A plasmacytoma, which is also a malignant clonal expansion of plasma cells, presents as a single lesion with similar imaging characteristics to the lesions of multiple myeloma (12).

Giant cell tumors are rare, benign, but locally aggressive lytic lesions of the bone. They usually occur around 20-30 years of age and are more common in Asian women (16). They usually occur in the skull and pelvic bones but can be seen in the vertebral body. On CT, it is lytic with thinning of the cortex and rarely (<5%) cause adjacent sclerosis. It is usually hypointense on T1 and heterogeneously hyperintense on T2 weighted imaging (13, 17).

TEACHING POINT

The top differential diagnosis for a newly diagnosed lytic bone lesion is typically metastatic malignancy in a 69-year-old patient. However, a lytic vertebral lesion in a patient who is status post aortic aneurysm repair may be due to an aortic wall to vertebral body fistula. This case is to highlight this possibility and educate radiologists to assess the integrity of the aortic wall and look for a possible connection between the aorta and the vertebral body in order to make the diagnosis and prevent the risk of hemorrhage and possible sepsis.

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FIGURES

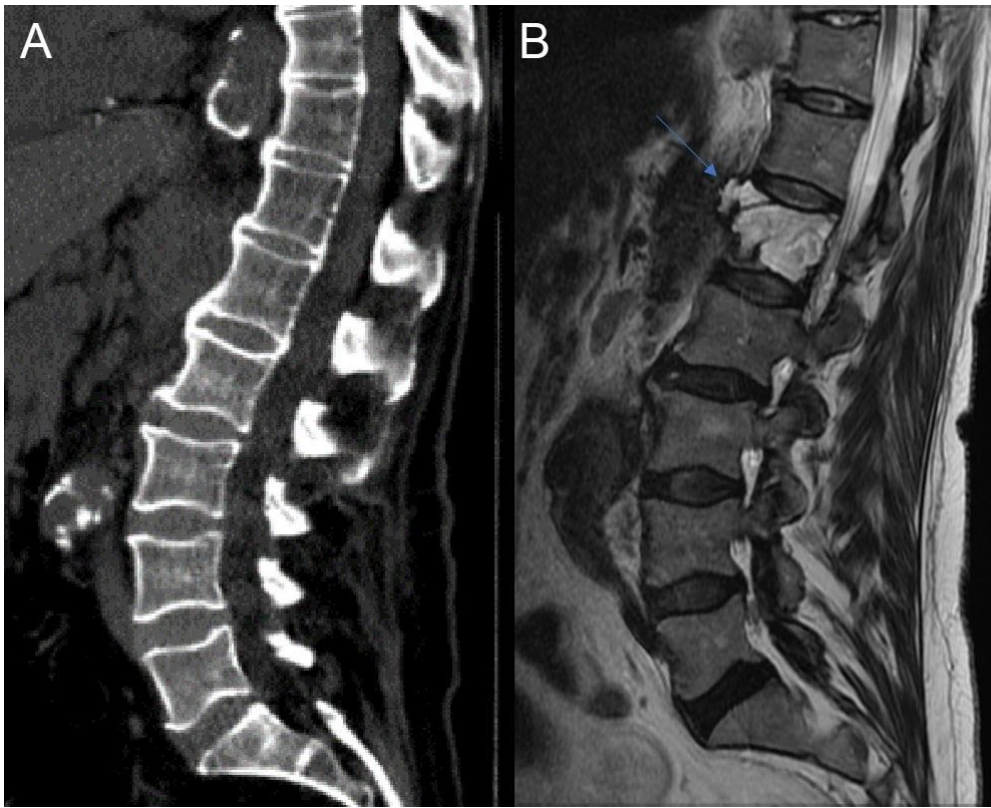


Figure 1: 69-year-old man with aortovertebral fistula.

A) Sagittal CT image 2 years prior to MRI demonstrates normal appearance of the L1 vertebral body.

B) Sagittal T2 weighted MRI without contrast shows fistulous connection between the wall of the abdominal aortic aneurysm and the L1 vertebral body (blue arrow). The vertebral body marrow signal is replaced with hyperintense T2 signal and shows internal trabeculations.



Figure 2 (left): 69-year-old man with aortovertebral fistula.

Sagittal T1 weighted MRI without contrast shows fistulous connection between the wall of the abdominal aortic aneurysm and the L1 vertebral body (blue arrow). The vertebral body marrow signal is replaced with intermediate T1 signal.

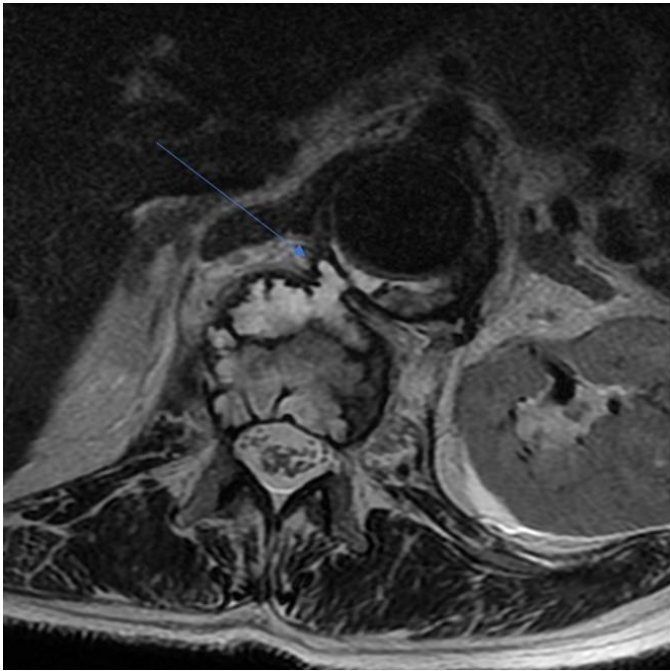


Figure 3: 69-year-old man with aortovertebral fistula. Axial T2 weighted MRI without contrast shows fistulous connection between the wall of the abdominal aortic aneurysm and the L1 vertebral body (blue arrow). The vertebral body marrow signal is replaced with hyperintense T2 signal and shows internal trabeculations.

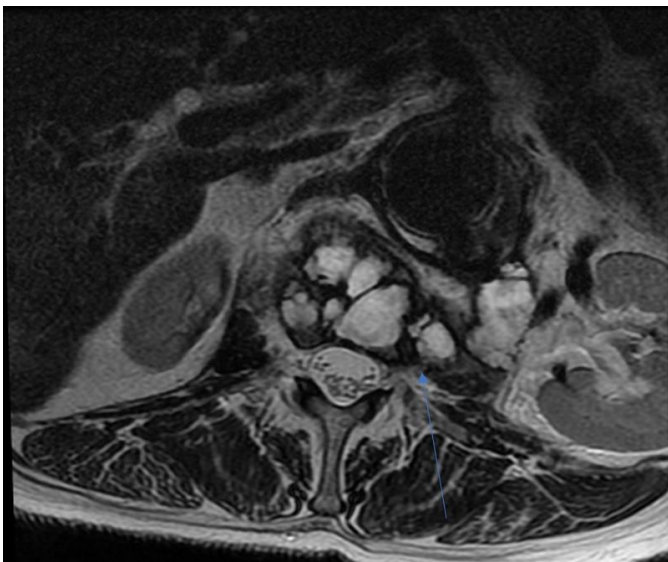


Figure 4: 69-year-old man with aortovertebral fistula. Axial T2 weighted MRI without contrast shows connection between the left lateral wall of the L1 vertebral body and a hyperintense T2 signal collection in the left psoas muscle (blue arrow). The vertebral body marrow signal is replaced with hyperintense T2 signal and shows internal trabeculations.



Figure 5: 69-year-old man with aortovertebral fistula. Sagittal CT angiogram image of the abdomen in the arterial phase after administration of 100 cc Visipaque 320 intravenous contrast. Image shows lucency of the L1 vertebral body with fistulous connection between the wall of the abdominal aortic aneurysm and the L1 vertebral body anteriorly (blue arrow).

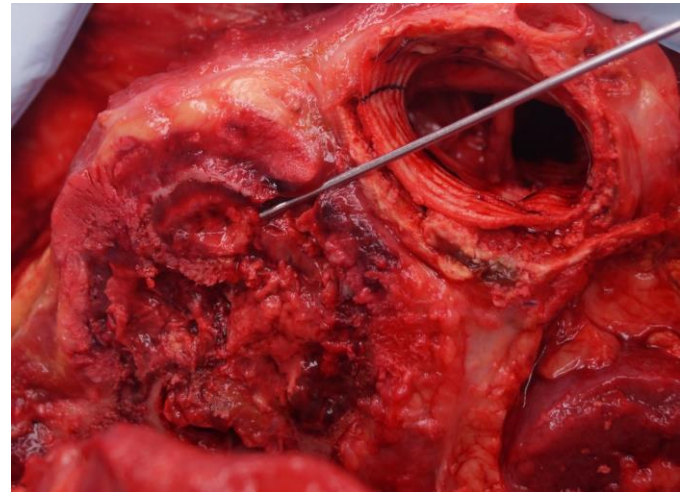


Figure 6: 69-year-old man with aortovertebral fistula. Axial CT angiogram image of the abdomen in the arterial phase after administration of 100 cc Visipaque 320 intravenous contrast. Image shows lucency of the L1 vertebral body with fistulous connection between the wall of the abdominal aortic aneurysm and the L1 vertebral body anteriorly (blue arrow).

Figure 8: 69-year-old man with aortovertebral fistula. Gross section demonstrating fistula tract between aortic graft and vertebral body (unfixed).

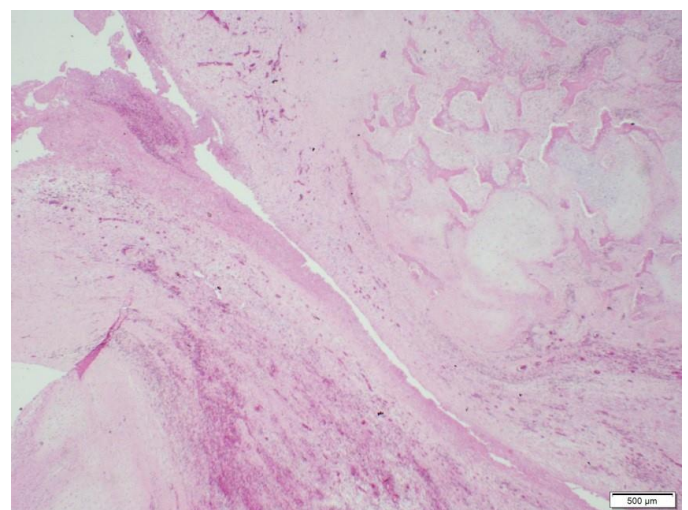
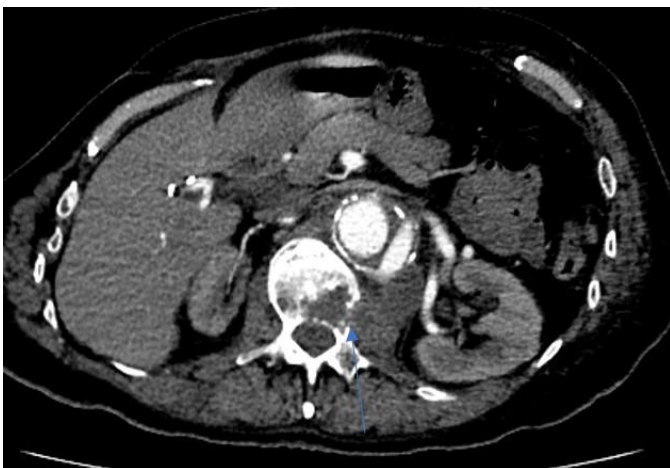


Figure 7: 69-year-old man with aortovertebral fistula. Axial CT angiogram image of the abdomen in the arterial phase after administration of 100 cc Visipaque 320 intravenous contrast. Image shows a connection between the left lateral wall of the L1 vertebral body and a low density collection in the left psoas muscle (blue arrow).

Figure 9: 69-year-old man with aortovertebral fistula. Microscopic examination demonstrating fistula tract between aortic graft and vertebral body. H&E, 2x

Etiology	Cystic medial degeneration and weakening of the wall
Prevalence	Estimated prevalence of asymptomatic TAAs is 0.16 percent
Gender Ratio	2-3 times more common in males than females
Age predilection	Peak incidence is around 60-70 years of age
Risk factors	Include but not limited to: 1. Bicuspid aortic valve 2. Connective tissue disorders such as: Marfan and Ehlers-Danlos Syndromes 3. Aortitis caused by multiple different factors 4. Same risk factors for atherosclerosis such as: hypertension, smoking and hypercholesterolemia
Treatment	Medical management: Smoking cessation, BP control Repair: Open vs Endovascular
Possible complications	Expansion, rupture, dissection, and fistula formation
Imaging Findings and modalities for diagnosis and screening	Imaging is important for diagnosis of enlarged lumen and subsequent surveillance once diagnosed. ECHO, MRI, and CT are all accepted modalities

Table 1: Summary table of Thoracic aortic aneurysms.

Differential Diagnosis	CT	MRI	NM
Lytic Metastatic disease: multiple myeloma, breast cancer, and lung cancer.	Lytic lesion with irregular margins with a possibly disrupted cortex	T1: Usually Hypointense. Enhances with contrast T2: Isointense to hyperintense on T2	PET/CT: MM is not PET active. Breast and lung cancer are PET avid.
Aneurysmal Bone Cyst	Lytic lesion with sharply defined thin sclerotic border	The periphery is hypointense on T1 and T2 with focal areas of T1 and T2 hyperintensity throughout lesion. Fluid-Fluid level with septations which may enhance	Bone scan: Increased uptake peripherally
Malignant hemangioma	Lytic lesion with dotted and linear areas on attenuation due to destruction of trabeculations and thickening of the remaining trabeculae	T1: Hyperintense Enhances with contrast T2: hyperintense	Bone scan: No increased uptake
Multiple myeloma/Plasmacytoma Bone Cyst	Punched out, lytic lesions, single for plasmacytoma and multiple for myeloma	T1: Hypointense to isointense T2: Isointense to hyperintense	PET/CT: Punched out lytic lesions which may show increased uptake or may be cold lesions
Giant Cell tumor	Lytic lesion without sclerotic border and thinned cortex	T1: Hypointensity of solid components which may enhance T2: Heterogenous areas of low signal	Bone scan: Increased uptake in the periphery on delayed images

Table 2: Differential diagnosis table for Aortovertebral fistula.

ABBREVIATIONS

ABC: Aneurysmal Bone Cyst
COPD: Chronic Obstructive Pulmonary Disease
CT: Computed Tomography
CTA: Computed Tomography Angiography
DM: Diabetes Mellitus
ECHO: Echocardiogram
HTN: Hypertension
L-spine: Lumbar spine
MRA: Magnetic Resonance Angiography
MRI: Magnetic Resonance Imaging
PET/CT: Positron Emission Tomography/Computed Tomography
TAA: Thoracic Aortic Aneurysm

KEYWORDS

Aortic aneurysm; fistula; endoleak; vertebral body hematoma; lytic spine lesion

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