Case Report

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Chronic contained ruptured abdominal aortic aneurysm with a rare presentation of lower limb neuropathic claudication: a report of two cases

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Introduction and importance: A vertebral body erosion that takes place due to a chronic contained rupture of an abdominal aortic aneurysm is an especially rare vascular pathology that comprises less than 5% of all causes of vertebral body erosion. Chronic contained rupture of an abdominal aortic aneurysms are primarily observed in hemodynamically stable patients whose chief complaint is lower limb neuropathic pain. This entity is extremely misleading and this results in delayed management of those patients increasing the morbidity and mortality rates.

Case presentation: We present the two cases of 62-year-old and 65-year-old males. Preoperative radiology for each patient showed an infrarenal aortic aneurysm with a retroperitoneal hematoma in contact with the lumbar vertebral bodies and psoas muscle. The draped aorta sign was evident in both cases.

Clinical discussion: A curative surgical intervention was accomplished for both patients, respectively. This was achieved through the removal of the existing hematomas that were compressing the vertebrae in addition to the complete isolation and resection of the respective abdominal aortic aneurysms along with thorough reconstruction of the aortoiliac spindles with patent synthetic grafts to ensure the patency of the preexisting vascular axis.

Conclusion: A contained rupture of an abdominal aortic aneurysm is a rare occurring vascular pathology that manifests with nonspecific symptoms, such as femoral neuropathy and lower back pain proportionate to the degree of the level of erosion of the affected lumbar vertebrae. This will increase the possibility of misdiagnosis and delays in treatment. Such a life-threatening vascular emergency should be timely detected and treated to avoid its complications and patient mortality.

Keywords: abdominal aortic aneurysm, case report, contained rupture of an abdominal aortic aneurysm, draped aorta sign, neuropathic claudication, vertebral body erosion

Introduction

An extremely rare subsequent event of a chronic contained rupture of an abdominal aortic aneurysm (CCR-AAA) is vertebral body erosion (VBE). It makes up less than 5% of all causes of VBE[1]. Patients with CCR-AAAs generally present with a stable hemodynamic status but nevertheless, complain of nonspecific lower back pain or lower limb neuropathic symptoms[2]. CCR-AAAs are characterized by the presence of pooling blood outside the lumen of the corresponding aortic aneurismal sac and into the retroperitoneal cavity. This is either manifested in a computed tomography scan or at the time of surgical intervention[3,4]. The aortic wall rupture symbolizes the present weakness of the aortic wall to sustain the occurring blood pressure. In the published literature, studies have demonstrated that the posterior arterial wall tear tends to be small in size and the subsequent pseudoaneurysm is chiefly sealed off with the surrounding anatomical structures, such as the vertebral bodies and the psoas muscle. This results in the overall stable hemodynamic state that the patients present with. In addition, the chronic bulging of the pseudoaneurysmal sac results in mechanical compression of these surrounding structures and leads to undefined lumbar vertebral erosion and lower back pain. This pain might radiate to the testes, back, inguinal region, or hip due to the direct irritation of the psoas muscle[5–8]. This rare

HIGHLIGHTS

- Chronic contained rupture of an abdominal aortic aneurysm is an extremely rare vascular pathology (< 100 cases worldwide).
- Symptoms are nonspecific. Therefore, misdiagnosis could occur and this delays the therapeutic surgical intervention.
- An extremely specific sign for this pathology is a radiological sign called: 'The draped aorta sign'.
- Complications are life-threatening. This highlights the importance of a timely surgical intervention.
- Surgical repair of the arterial wall defect and removal of the hematoma are the gold standard therapeutic approach.

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clinical presentation of such a pathology (< 100 documented cases worldwide) renders making a diagnosis a difficult endeavor and this results in potentially life-threatening complications mostly due to the freeing of the anatomically contained hematoma[9]. A Computed Tomography scan with contrast enhances the ability to screen, diagnose, and prepare timely surgical interventions. Treatment of CCR-AAAs is accomplished through various surgical forms, such as open surgical or endovascular surgical repair where the primary objective is to reconstruct the damaged aortic segment and remove the resultant pseudoaneurysm[3,4].

The work has been reported in line with the Surgical CAse REport (SCARE) criteria and the revised 2020 SCARE guidelines[10].

Presentation of case

Patient information

We present the two cases of a 62-year-old male (Patient A) and a 65-year-old male (Patient B). Patient A has a known history of arterial hypertension while patient B was previously healthy. Both patients were referred to our university hospital's Vascular Surgery Clinic with the chief complaint of chronic lower back pain. The pain began 6 months prior to admission, it was sharp, constant, scaled 7–8/10 on the patients' numerical pain scale, radiated to the posterior thigh and lateral aspect of the thighs, was aggravated by positions that elevate the abdominal pressure (cough and hip motions), and was, at the early stages, partially relieved by over-the-counter analgesics while at the later stages, became unresponsive to them. The symptoms of both patients were associated with palpable pulsatile periumbilical masses that began to be felt simultaneously with the onset of the pain. Both patients denied any occurring coldness, pallor, cyanosis, or lower limb swelling. Additionally, both patients denied any history of immune diseases, recent infections, or trauma to the back or abdomen. Patient A's drug history involved oral antihypertensive medications, whereas patient B's drug history was negative. Both patients' psychosocial histories only included a smoking history of 2-pack-years and 3-pack-years, for patients A and B, respectively. Furthermore, both patients' surgical, family, and allergic histories were unremarkable. Patient A's and patient B's BMI were 27 and 29 Kg/m^2, respectively.

Clinical findings

A physical examination revealed acceptable vital signs according to both patients' ages. Both patients demonstrated pallor upon inspection. Furthermore, both of them had a pulsatile periumbilical abdominal mass upon palpation. The pulses along the arterial axes were normal in both patients' lower limbs, and the auscultation revealed localized bruit over the site of the previously mentioned masses. Other physical examination results were unremarkable.

Diagnostic assessment

Both patient A and patient B had similar radiological imaging results and they are summarized as follows: By performing the Duplex Ultrasound, an infrarenal aortic aneurysm measuring (73 × 70 mm) in patient A, whereas the same one by patient B was found measuring (82 × 80 mm). Both aneurysms were accompanied by arterial wall thrombosis while no other aneurysms were detected. Further analysis was achieved by performing a high-resolution contrast-enhanced Multi-Slice Computed Tomography (MSCT) scan of the abdomen and pelvis. In each of our patients, it revealed an infrarenal aortic aneurysm with a retroperitoneal lesion consistent with a hematoma formation. Additionally, these hematoma formations appeared to be infiltrating the lumbar vertebral bodies and psoas muscle. The diminished line between the posterior aortic wall and the vertebral column is a specific sign for CCR-AAA and is called the draped aorta sign. Furthermore, smooth and well-corticated vertebral body deteriorations with callous borders were visualized. These findings are known to be caused by the existing aneurysms due to chronic recurrent arterial pulsations. Moreover, we also noted cloudiness of the iliopsoas contours, which were likely to be caused by small vascular leaks. (Fig. 1 A, B) for patient A and (Fig. 1 C, D) for patient B. The remaining arterial tree was found to be normal with no remarkable findings. A conclusive laboratory panel was requested. The only anomaly noted was the hemoglobin value of patient A. It was 10 g/dl, whereas it was 9 g/dl for patient B. Other laboratory markers including infection parameters were all within normal values.

Given the previous clinical picture for both patients, surgery was the chosen treatment approach. Both patients were assigned a nil-per-mouth nutrition status prior to surgery, proper intravenous access was established, and suitable preoperative antibiotics were administered. No challenges or deviations from the treatment plan were faced in any of the perioperative periods.

Therapeutic intervention

Both surgical interventions for patient A and patient B were successfully achieved in our tertiary university hospital where both interventions were conducted under general anesthesia with no reported perioperative complications. Both operations were accomplished by a Vascular Surgery Specialist with 15 years of experience. The surgical technique and details of both operations are summarized as follows: the operations were initiated by performing a longitudinal midline abdominal incision to achieve optimal surgical exposure of the field. We found no thickening of the arterial wall or retroperitoneal adhesions. However, a retroperitoneal hematoma was demonstrated. A portion of the abdominal aortic aneurysm was firmly attached to the surrounding soft tissue. The aorta was cross-clamped below the level of the renal artery and the aneurysm was then cut open. A partial defect in the aneurismal posterior wall was observed at the position where the preoperative contrast-enhanced MSCT scan suggested its presence Figure 2 (A, B). The aorta was trimmed below the level of the renal artery and a Y-shaped graft replacement was achieved (16 × 8 mm DACRON). The distal side was anastomosed to the common femoral artery on both sides. Symptoms of both patients subsided postoperatively and they both underwent complete recovery. Proper postoperative care included sterile wound dressings, intravenous fluid support, and postoperative antibiotics and analgesia. Both patients were discharged on the seventh postoperative day and have been followed up in our specialized hospital clinic for 2 years thus far. During the postoperative clinical assessments, the results of the physical and radiological exams for both patients were within normal. Nowadays, they are assigned for annual clinical visits as long-term follow-ups.

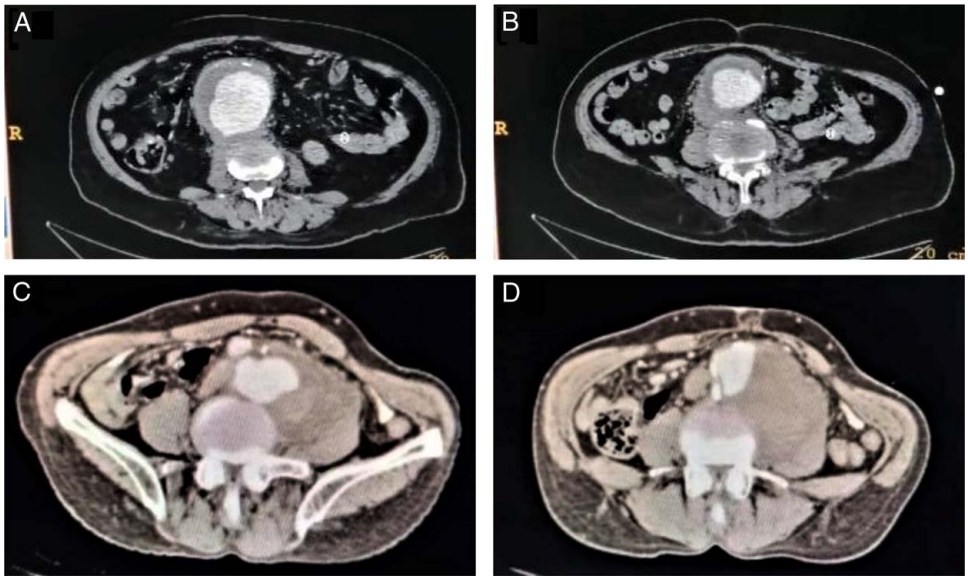

Figure 1. A: Preoperative contrast-enhanced Multi-Slice Computed Tomography (MSCT) image of the abdomen and pelvis of patient A. It revealed a chronic contained rupture of an infrarenal abdominal aortic aneurysm with posterolateral retroperitoneal hematoma extending over the bony vertebral edge (Draped aorta sign). B: Preoperative contrast-enhanced MSCT image of the abdomen and pelvis of patient A. It revealed a chronic contained rupture of an infrarenal abdominal aortic aneurysm with posterolateral retroperitoneal hematoma extending over the bony vertebral edge (Draped aorta sign). In addition to a well-corticated vertebral body destruction. (C, D): Preoperative contrast-enhanced MSCT image for the abdomen and pelvis of patient B. It revealed a chronic contained rupture of an infrarenal abdominal aortic aneurysm with a left posterolateral retroperitoneal hematoma extending over the psoas muscle and the bony vertebral edge (Draped aorta sign).

Discussion

CCR-AAAs were first depicted by Katz *et al.* in 1962. Less than 100 documented cases exist since then[1,2]. This rare vascular pathology is characterized by the presence of blood outside the lumen of the aneurismal sac of the aorta and leaking into the retroperitoneal cavity[2]. CCR-AAAs comprise 2.7–4% of all surgical infrarenal abdominal aortic aneurysms and they are predominantly occurring in males with an average age of incidence of 73.1 ± 8.3 years[11]. CCR-AAAs comprise less than 5% of all incidences of VBEs where the most frequently affected aortic segment is the infrarenal segment. Most affected patients are hemodynamically stable at the time of presentation. Several hypotheses were stipulated to explain these phenomena, such as normal heart rates, normal/controlled arterial hypertension, and small-sized tears in the aortic wall that could be sealed off by the hematoma itself[5,6]. The resulting retroperitoneal hematomas have a pulsatile effect that plays an important role in causing repetitive physical irritation and pressure that result in subsequent bone ischemia and vertebral lysis and deterioration[12]. Affected patients typically present with stable vital signs and nonspecific clinical symptoms, such as lower back pain (64%), pain in the groin region (14%), and femoral neuropathy (8%). Furthermore, most abdominal aortic aneurysms are chiefly asymptomatic but nevertheless, they can be manifested as a vague abdominal pain until the point of aneurismal wall rupture. Nonetheless, abdominal aortic aneurysms could become a CCR-AAA as soon as contained bleeding is present[3]. During clinical examination of these patients, physical examination mostly yields

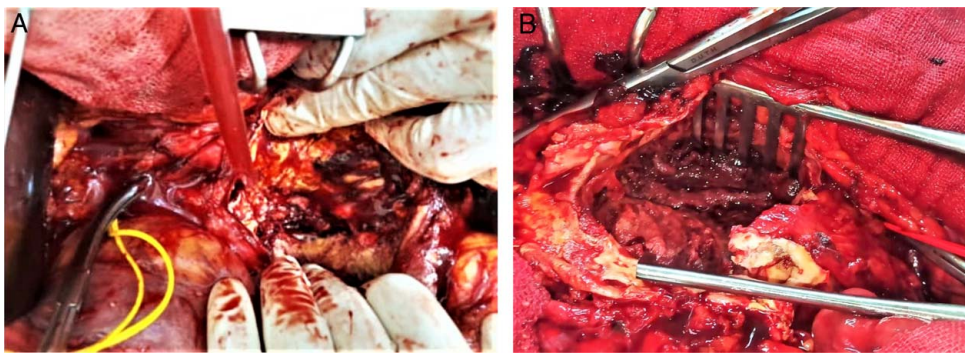

Figure 2. A: Intraoperative image from the operation on patient A. It demonstrates the chronic contained ruptured abdominal aortic aneurysm. It demonstrates the (2 × 2 cm) punched-out aortic wall defect located on the posterior wall of the aneurysm. Through this defect, the deteriorated vertebral body was discovered. B: Intraoperative image from the operation on patient B. It demonstrates the chronic contained ruptured abdominal aortic aneurysm.

normal vital signs with no hints toward acute distress of changes in hemoglobin levels. However, the patients' chief clinical symptom at the point of aneurismal rupture is an acute occurring, progressive, and sharp lower back pain that becomes constant and hindering. With regard to the utilized preoperative diagnostic modalities, an enhanced MSCT scan with/or without intravenous contrast is a suitable noninvasive radiologic assessment tool that aids in establishing a diagnosis of CCR-AAAs[13]. A pathognomonic radiological sign to suspect and diagnose CCR-AAAs is the draped aorta sign. It was initially demarcated by Halliday *et al.* and it describes a particular line between the posterior aortic wall and the adjacent structures that cannot be vividly identified[14,15]. In addition, smooth and well-demarcated vertebral erosions with sclerotic borders are considered auxiliary radiological findings that occur as a result of chronic recurrent erosions due to arterial pulsation. Furthermore, blurring of the iliopsoas borders that are potentially caused by minor vascular leakage was also reported in radiology[14]. Possible differential diagnoses for vertebral body lysis include inflammatory and infective etiologies[16]. Based on the previous clinical and radiological merits, open surgical intervention is the current gold standard treatment option for CCR-AAAs. This method has proven its potency in limiting potential life-threatening complications, such as arterial wall rupture and progressive vertebral degradation[17]. As for the prognosis of CCR-AAAs, if they are timely diagnosed and treated, the prognosis is favorable for the affected patients. Otherwise, fatal results could take place[9].

Conclusion

A contained rupture of an abdominal aortic aneurysm is a markedly rare occurring vascular pathology that manifests with misleading symptoms, such as lower back pain and femoral neuropathy consistent with the degree of the level of erosion of the affected lumbar vertebrae. This, in turn, leads to an increased chance of misdiagnosis and delays in treatment. Life-threatening vascular entities such as this one should be detected and treated in due time to avoid the ensuing complications and patient mortality. To the best of our knowledge and based upon the review of the published literature, we believe no previous documented cases from our country discussing such a vascular pathology, exist. This highlights the value and importance of our findings.

Ethics approval and consent to participate

Institutional review board approval is not required for deidentified single case reports or histories based on institutional policies.

Consent of patient

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

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Author contribution

O.A. and O.H.: Conceptualization, resources, methodology, data curation, investigation, who wrote, original drafted, edited, visualized, validated, and literature reviewed the manuscript.

A.M.: Vascular Surgery specialist who performed and supervised the operation, in addition to supervision, project administration, resources, and review of the manuscript.

O.A.: The corresponding author who submitted the paper for publication. All authors read and approved the final manuscript.

Conflicts of interest disclosure

The authors declare that they have no financial conflict of interest with regard to the content of this report.

Research registration unique identifying number (UIN)

N/A.

Guarantor

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Availability of data and materials

The datasets generated during and/or analyzed during the current study are not publicly available because the Data were obtained from the hospital computer-based in-house system. Data are available from the corresponding author upon reasonable request.

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