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CASE REPORT

# Ruptured Aneurysm of the Common Iliac Artery Caused by *Brucella melitensis*: A Case Report

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**Introduction:** *Brucella* is a genus of aerobic Gram negative bacteria that causes the disease brucellosis. It is considered a zoonotic infection transmitted to humans by ingestion of unpasteurised dairy products. Although aortic involvement is rarely seen, it can be a life threatening complication of this disease. This case report describes a ruptured aneurysm of the common iliac artery (CIA) due to secondary infection by *Brucella melitensis.*
**Report:** A 79 year old man with a known isolated aneurysm of the CIA presented with acute abdominal pain. Contrast enhanced computed tomography (CT) revealed rupture of the aneurysm. The patient underwent prompt endovascular repair. Several weeks after an uneventful recovery, the patient presented with spiking fever and abdominal discomfort. CT revealed an abscess anterior to the CIA. Blood and pus cultures grew *B. melitensis*. In recurrent re-admissions, conservative antibiotic therapy proved to be insufficient. Eventually, neo-aorto-iliac system (NAIS) reconstruction using bilateral femoral veins was performed to provide definitive treatment four months after initial presentation.
**Conclusion:** Although *Brucella* infected aneurysms are rare, they are associated with life threatening disease. Diagnosing this type of brucellar infection can be challenging owing to the long incubation time needed for blood and tissue cultures. Definitive treatment of these aneurysms often needs open surgery and antibiotics for complete treatment. Vigilant surveillance is required to monitor for post-operative complications such as graft infection, recurrent (false) aneurysm, and abscess formation.

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## INTRODUCTION

Brucellosis is a zoonotic disease transmitted to humans by the consumption of infected, unpasteurised dairy products.[1] It has a global distribution and is endemic in most countries in the Middle East. Symptoms include insidious onset of fever, malaise, night sweats, and arthralgias. The disease is caused by *Brucella* spp., which are Gram negative, intracellular, immobile, and non-encapsulated bacteria.[1] Vascular manifestations of *Brucella* infections have a prevalence of 3%, of which the vast majority include endocarditis.[1] In cases of aortic involvement, brucellosis typically manifests as an infected aneurysm.[2,3] Although these cases are reported, it is a very unusual presentation of the disease.

Definitive treatment of infected aneurysms must consist of a combination of antibiotic therapy and vascular surgery. The gold standard in these cases is open surgery with aneurysmectomy, local debridement, and graft replacement.[4] For an infected aorto-iliac aneurysm, a neo-aorto-iliac system (NAIS) reconstruction (autogenous venous graft) can be used.[5] Because these grafts are placed in an infected surgical site, post-operative complications such as graft infection and recurrent (false) aneurysm can occur and should be watched for vigilantly.[6] Even though the incidence of infected aneurysms is low, they are associated with significant morbidity and mortality, mainly due to a high rate of rupture.

In this case report, the rupture of an infected isolated aneurysm of the right common iliac artery (CIA) by *Brucella melitensis* and subsequent treatment is described.

## REPORT

A 79 year old Turkish man with a history of an abdominal aneurysm (33 mm) and isolated right and left CIA aneurysms (31 mm and 27 mm respectively on last follow up eight months prior to admission) presented to the emergency department (ED) with acute and progressive abdominal pain. There was no recent history of abdominal

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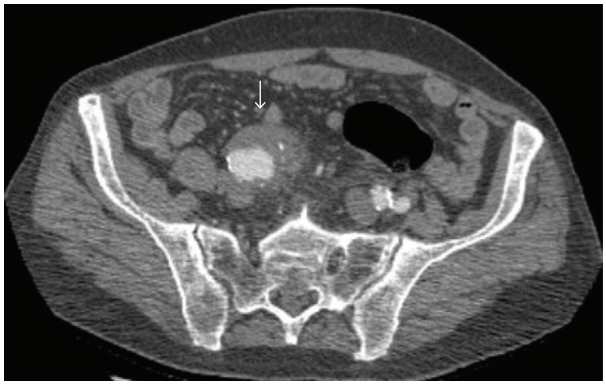

**Figure 1.** Contrast enhanced computed tomography. Axial image of the contained rupture of the right common iliac artery aneurysm (40 mm, indicated by the arrow). The aneurysm is saccular and the arterial wall seems hazy, which make it, in retrospect, suspicious of infection.

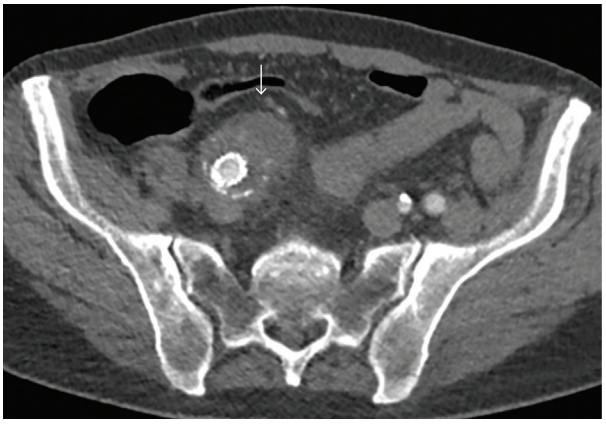

**Figure 2.** Contrast enhanced computed tomography. Axial image of the soft tissue mass (25 × 37 mm, indicated by the arrow) anterior to the common iliac artery during the second hospital admission.

trauma or surgery. The patient was conscious, with normal vital parameters, and had no fever. Abdominal examination revealed diffuse tenderness on palpation. Laboratory tests showed a haemoglobin level of 11.4 g/dL (reference interval [RI] 13.8—17.2 g/dL), leucocytes of 8.2 mmol/L (RI 4.5—11.0 mmol/L), and a C reactive protein (CRP) level of 37 mg/L (normal <10 mg/L). Computed tomography (CT) showed a (contained) ruptured aneurysm of the CIA with a maximum transverse diameter of 40 mm (Fig. 1, Video S1 [Supplementary Material]).

The patient was immediately transferred to a hybrid operating room, where percutaneous endovascular repair was performed. The right internal iliac artery was embolised with 10 mm coils, followed by infrarenal endovascular aneurysm repair (EVAR; C3 Gore® Excluder® [W.L. Gore and Associates, Flagstaff, AZ, USA]) extending into the right proximal external iliac artery. Completion angiography showed successful exclusion of the ruptured aneurysm (Video S2; see Supplementary Material). The patient recovered uneventfully and was discharged home after three days.

Two weeks after surgery, the patient complained of increasing pain in the groin area on the right side. His vital functions were normal (blood pressure 113/60 mmHg, heart rate 77 beats/minute) and he had no fever (temperature 36.8oC). Laboratory results showed a CRP level of 91 mg/L. CT was performed, which showed a soft tissue mass of 25 × 37 mm anterior to the right CIA (Fig. 2, Video S3 [Supplementary Material]). The differential diagnosis included abscess formation or (infected) haematoma. Percutaneous aspiration of the mass was performed, which yielded old haemorrhagic fluid. Because of a suspected haematoma infection, two blood cultures were taken and intravenous antibiotics were administered for two weeks (flucloxacillin 1 000 mg four times daily). The infectious parameters increased (CRP level 181 mg/L), but both blood and aspiration specimen cultures showed no growth of micro-organisms. After antibiotic treatment, CRP levels decreased to 130 mg/L and the patient was discharged from the hospital in good clinical health.

Seven weeks after surgery, the patient presented again with fever, nausea, and mild abdominal pain. Laboratory tests showed a CRP level of 65 mg/L. A new CT scan was performed, which showed progression of the mass (26 × 45 mm) anterior to the CIA, this time highly suspicious of an abscess owing to its confined shape (Fig. 3, Video S4 [Supplementary Material]). Percutaneous drainage was performed and the pus obtained was cultured again. Two blood cultures were taken and broad spectrum intravenous antibiotics (cefuroxime and metronidazole) were commenced. After a few days, both blood and pus cultures showed growth of *B. melitensis*. The antibiotics were switched to gentamicin intravenously and doxycycline orally. After two weeks of intravenous treatment and multiple percutaneous drainages, the abscess had significantly shrunk in size and the infection parameters had decreased (CRP level from 152 mg/L to 48 mg/L). Ultrasound of the abdomen showed decreasing abscess diameters. Echocardiography showed no signs of endocarditis. As the patient was hesitant about open surgery, he was discharged with oral antibiotics consisting of doxycycline and rifampicin with the intention of lifelong treatment.

Despite continued treatment with antibiotics, the general malaise and weight loss progressed four months after initial presentation. Definitive treatment (by NAIS reconstruction)[5] took place in an academic hospital. Bilateral femoral veins were harvested (25 cm both sides) and proximal side to side anastomosis was performed to create a bifurcated graft. When entering the operation area, infected tissue around the right CIA was seen without evidence of pus. A small affected aortic segment was resected. There was no back bleeding from the embolised internal iliac artery. Then, the aorta was clamped suprarenally (30 minutes) in order to remove the EVAR and insert the proximal anastomosis just inferior to the renal arteries. Subsequently, the distal anastomoses were made in the proximal external iliac artery on the right side and the origin of the proximal CIA on the left side. The hypogastric coils were left in place. The total duration of surgery

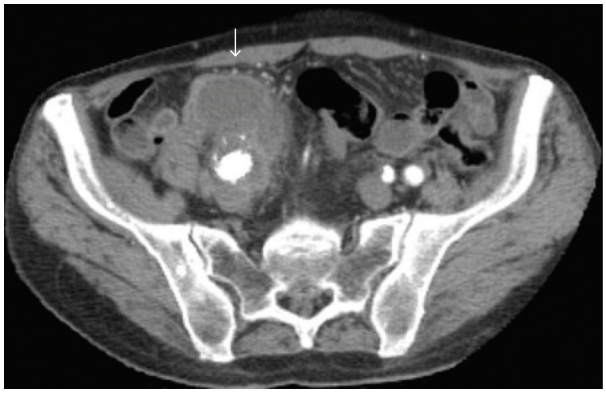

**Figure 3.** Contrast enhanced computed tomography. Axial image of the progression of the soft tissue mass (26 × 45 mm, indicated by the arrow) anterior to the common iliac artery during the third hospital admission.

was approximately 10 hours. A few hours after the procedure, re-laparotomy was needed for post-operative bleeding caused by diffuse oozing. The abdomen was packed with gauzes, which were successfully removed two days later. After six days in the intensive care unit and one month on the surgical ward, the patient was referred to a nursing home and treated with oral antibiotics (doxycycline/co-trimoxazole) for another month. Approximately one year post-operatively, the patient has fully recovered with no signs of recurrent infection. Follow up CT showed no procedural complications (Video S5; Supplementary Material). Fluorodeoxyglucose positron emission tomography CT scan revealed complete remission of the inflammatory reaction.

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.

## DISCUSSION

This case report describes the rupture of an isolated iliac aneurysm due to secondary infection by *B. melitensis.* An infected aneurysm by *Brucella* spp. is a rare finding. In total, only 49 infected aorto-iliac aneurysms have been published in the literature (Table S1, Appendix S1 [Supplementary Material]). However, to the best of the authors' knowledge, no other case report has mentioned an infected isolated aneurysm of the CIA by this pathogen.

*Brucella* is a rare pathogen in Western Europe. It mainly affects populations living in rural areas in Mediterranean, Middle East, and Latin American countries.[1] The patient discussed in this report was probably exposed during a family visit to Turkey. This visit was approximately six months before his initial presentation to the ED. As *Brucella* spp. can act as prolonged low grade infections, this scenario of contamination is certainly plausible.[1]

Furthermore, in this case, the diagnosis was missed initially due to negative blood and specimen cultures. From previous research, it became clear that the detection of *Brucella* spp. in blood cultures is difficult, as these bacteria have a long incubation time of up to 10 days.[7] Routinely, blood cultures are discarded after 5—7 days, so isolation of slow growing Brucellae can be missed easily.

Infected aneurysms caused by *Brucella* spp. are associated with high morbidity and mortality rates (21%).[8] Owing to the low number of reported cases, recommendations for treatment have not been established. General principles of treatment consist of a combination of antibiotic and surgical therapy.[4] Antibiotic treatment for *Brucella* spp. aims to create acidic intracellular environments, which can be achieved with aminoglycosides, doxycycline, and rifampicin. Combination therapy of two or more of these antibiotics together with prolonged duration of treatment are necessary for full eradication of the bacteria. Aminoglycosides should be administered intravenously for at least two weeks.[9] The main surgical aim depends on the type of surgery chosen. In general, endovascular therapy is useful as an initial treatment to achieve haemodynamic stability in a life threatening presentation until definitive reconstruction can be performed (bridge to surgery).[4] If an open procedure is performed, the general principles of vascular graft infection can be followed. This includes wide debridement of infected tissue, obtaining specimens for culture and arterial reconstruction.

In this case, there was a low suspicion of infection initially and endovascular surgery was chosen as treatment. Eventually, this resulted in graft infection and abscess formation. As the patient did not want invasive (open) surgery at first, conservative therapy with lifelong antibiotic treatment was attempted. Due to medical deterioration, open surgery became unavoidable and was performed four months after presentation.

## CONCLUSION

Brucellosis as the cause of an infected aneurysm is very rare. The diagnosis can be challenging owing to the long incubation time of blood and tissue cultures. There should be awareness of this disease in patients who live in endemic areas or consume unpasteurised dairy products. Definitive treatment by open surgery with arterial reconstruction needs to be performed for full control of the infection.

## ACKNOWLEDGEMENTS

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## APPENDIX B. SUPPLEMENTARY DATA

Supplementary data related to this article can be found at https://doi.org/10.1016/j.ejvsvf.2021.06.011.

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