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CASE REPORT

Lethal hemorrhage from a ureteric–arterial–enteric fistula diagnosed by postmortem CT angiography

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Abstract Fistula formation following pelvic surgery and radiotherapy, including ureteric–arterial fistulas (UAF), is well documented, however, ureteric–arterial–enteric fistula is extremely rare. Conventional autopsy is usually required for the definitive diagnosis of pelvic vascular fistulas although an accurate diagnosis can still be complicated and challenging. The role of post-mortem computed tomography (PMCT) as an adjunct to conventional autopsy is well documented in the literature. One of the limitations of PMCT is the diagnosis of vascular conditions. Post-mortem computed tomography angiography (PMCTA) is a recently introduced technique that can assist in detecting such pathology. We present a case of post-radiotherapy ureteric–arterial–enteric fistula presenting as massive rectal and vaginal bleeding diagnosed prior to autopsy on PMCTA. The role of PMCTA in the diagnosis of such a UAF has not previously been reported in the literature.

Keywords Ureteric–arterial–enteric fistula · Post-mortem computed tomography (PMCT) · Post-mortem computed tomography angiography (PMCTA) · Conventional autopsy

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Introduction

Fistula formation following pelvic surgery and radiotherapy, including ureteric–arterial fistulas (UAF), is well documented, however, c fistula is extremely rare. UAF is a difficult entity to detect in life and is associated with a high mortality rate. Conventional autopsy is usually required for the definitive diagnosis of pelvic vascular fistulas. The role of post-mortem computed tomography angiography (PMCTA) in the diagnosis of such cases has not previously been reported in the literature. We describe the PMCTA findings in UAF and discuss how this technique can be used to assist the forensic pathologist.

Case report

The deceased was a 76-year-old female diagnosed with endometrial carcinoma in 1995. She underwent total abdominal hysterectomy and bilateral salpingo-oophorectomy followed by radiotherapy. Years later she developed a presumed post-radiation fistula between the bladder and vagina. As a result she became incontinent with frequent episodes of urinary sepsis including *Pseudomonas* and *Candida*. She also had bilateral benign ureteric strictures leading to chronic renal impairment and requiring permanent ureteric stents.

Four weeks prior to her death the deceased experienced intermittent, small volume, vaginal bleeding. She was admitted to hospital but the exact cause of bleeding could not be found. There was no history of recent falls or trauma, no recent surgery and no thrombogenic medications. She was considered to be too frail for operative intervention. The bleeding eventually stopped and she was discharged. A few days later she revisited the emergency department with signs and symptoms of urinary tract

infection. She was also hypotensive and anemic but there was no apparent vaginal bleeding at that time. The following day she was admitted to the intensive care unit (ICU) for treatment of sepsis.

In the ICU she had uncontrollable pelvic bleeding, either from the vagina or rectum, causing her vital signs to deteriorate rapidly. An urgent contrast-enhanced computed tomography (CT) of the abdomen and pelvis was organized and dilute radiographic contrast media injected into the nasogastric tube as a part of the preparation for that procedure. While waiting for CT she collapsed with more pelvic hemorrhage. Full cardio-pulmonary resuscitative measures including external cardiac massage proved futile and she succumbed about an hour later.

Radiological findings

A whole body post-mortem CT scan (PMCT) was performed on the following day at our institute using a mortuary located Toshiba Aquilion, 16 multi-detector scanner (Toshiba Medical Systems Corporation, Tochigi, Japan). CT scanning of the deceased is an established technique performed on all individuals admitted to our institute over the last 6 years. It is primarily used to assist pathologists in determining cause and manner of death. Images were reviewed on a TeraRecon Inc AquariusNet workstation V4.4.1.4 (Foster City, CA 94404, USA) by the assigned pathologist and a forensic radiologist.

PMCT scan of the abdomen and pelvis revealed an irregular mass in the right side of the pelvic cavity. There was no clear fat plane between this mass and the distal right ureter (as outlined by an opaque stent), surrounding bowel and the right iliac vessels including a calcified iliac artery (Fig. 1). Despite the presence of a right ureteric stent, there was gross right-sided hydronephrosis and hydroureter. The bladder was not well visualized. A left ureteric stent was also in situ and the left kidney appeared small with irregular margins in keeping with chronic parenchymal disease. Diluted contrast was present in the stomach and small bowel having a mean Hounsfield unit (HU) density of 200–300. No free fluid or air was detected in the peritoneal cavity. The remainder of the abdominal organs were unremarkable and there was no obvious abdominal lymphadenopathy. Multiple small bilateral lung nodules were seen together with bilateral, symmetrical anterior rib fractures from 2 to 6, consistent with external cardiac massage.

In view of the circumstances surrounding death and preliminary CT findings suggesting a mass involving the right iliac vessels, PMCTA was performed 2 days after death. The right femoral artery and vein were individually cannulated and 1,700 ml of a solution containing Iosovue 370 mg/ml (Bracco s.p.a, Milan, Italy) mixed with polyethylene glycol 200 (PEG200, Merck Schuchardt OHG, Hohenbrunn, Germany) in a 1:10 ratio was infused into the artery using an embalming pump (Dodge, MA, USA). A whole body CT was done immediately after completion of the infusion.

Contrast leak from the right external iliac artery was identified entering into the pelvic mass (Fig. 2) and right ureter with retrograde filling of the right renal pelvis. Contrast was also noted in the contracted bladder flowing into the vagina and then externally during the infusion process. High density contrast fluid was also detected in the small intestine surrounding the mass which appeared to be denser in the post-angiographic scans when compared with the pre-angiographic CT images. We compared the HU measurement at a few locations within the small bowels in both the pre-angiographic scans and the post-angiographic scans and noted an increased in the mean HU from 200 to 300 (Fig. 3a) to 600–700 HU (Fig. 3b). This finding increased the suspicion of a possible communication between the artery, mass and small intestine. Minimal free contrast media was also noted in the peritoneal cavity.

Autopsy findings

A full body autopsy was performed on the following day. A large fibrotic and partly necrotic mass was identified in the right pelvic wall, which involved the urinary bladder, distal right ureter and a portion of the small intestine. The necrotic mass eroded through the wall of the bladder and vaginal, distal right ureter, adjacent small bowel and extended into the medial side of the right external iliac artery (Fig. 4). Contrast solution was also noted within the lumen of the surrounding small intestine.

Histological examination of the necrotic mass revealed extensive fibrosis and necrosis, acute inflammation, scattered hemosiderin-laden macrophages and areas of abscess formation. The fibroblasts showed marked cytological atypia, characterized by nuclear enlargement and smudged chromatin, consistent with radiotherapy effect. There was no evidence of residual or recurrent endometrial carcinoma of the sampled sections of the pelvic mass. There was however, a single 3 cm metastasis in the left lower lobe of the lung. Histological examination showed poorly differentiated endometriod adenocarcinoma, composed of highly atypical cells with marked nuclear atypia, arranged in solid sheets and poorly formed glands. Numerous mitoses and areas of tumor necrosis were also present.

Discussion

Ureteric–arterial fistulas (UAFs) [1–3] and enteric-arterial fistulas [4] are rare clinical occurrences. Ureteric–arterial–

Fig. 1 Pre-angiogram PMCT scan of the abdomen and pelvis in thick slab multiplanar reconstruction (MPR) view showing the mass the pelvic cavity (*white star*),the distal right ureter as outlined by an opaque stent (*thin white arrow*), surrounding bowel (*thick white arrow*) and the calcified *right* iliac vessels (*white arrow head*)

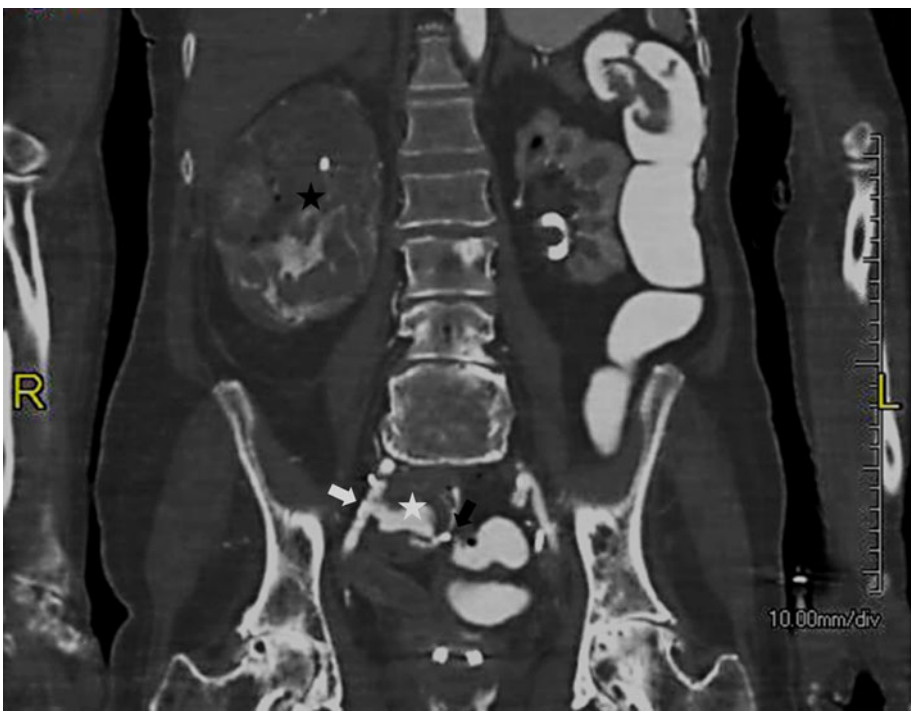

Fig. 2 Post-angiogram PMCT scan of the abdomen and pelvis in *coronal view* showing contrast leak from the *right* external iliac artery (*white arrow*) entering into the pelvic mass (*white star*) and *right* ureter with retrograde filling of the dilated *right* renal pelvis (*black star*) and adjacent small bowel (*black arrow*)

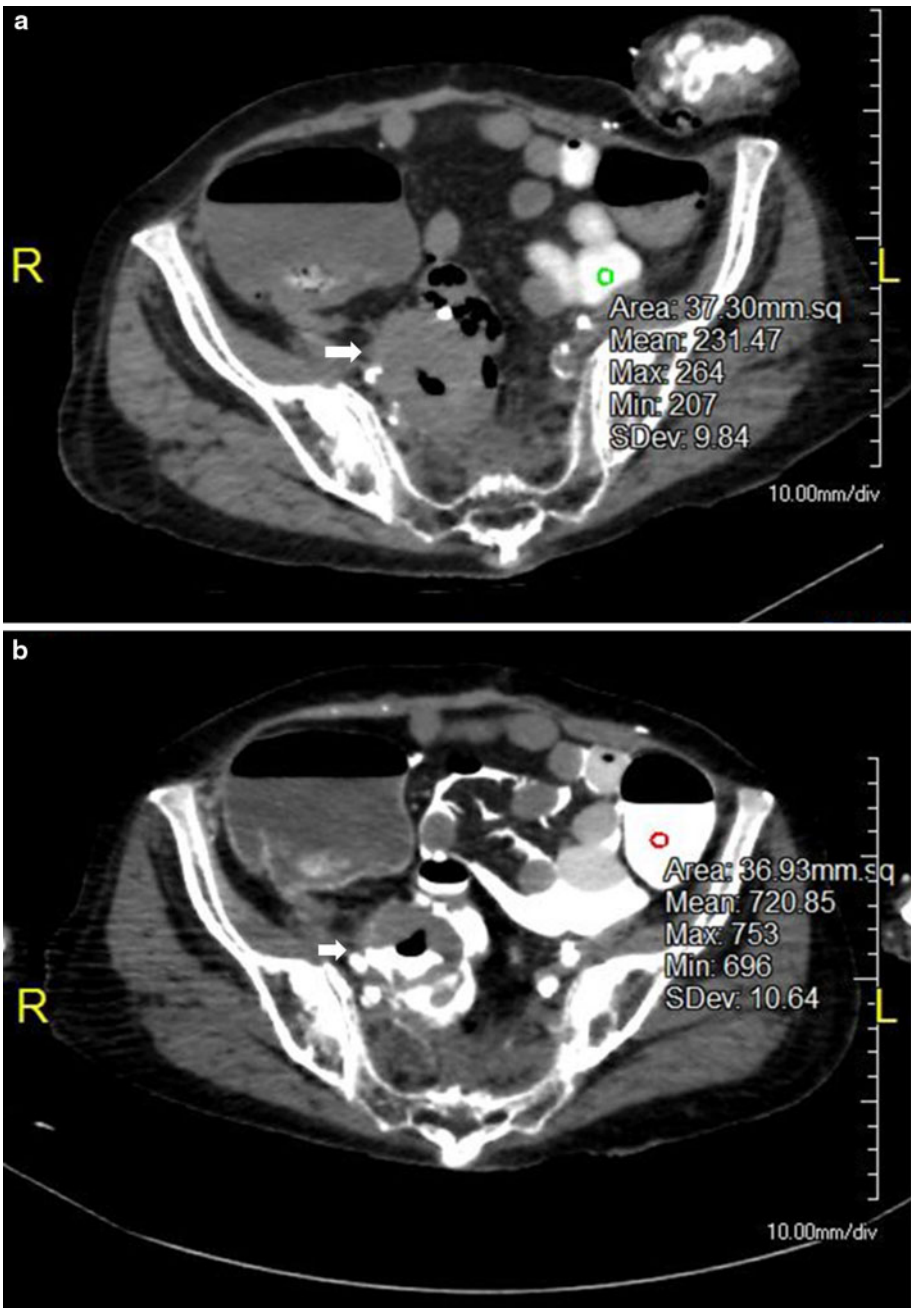

Fig. 3 CT scans of the abdomen and pelvis in *axial plane* showing the high density contrast fluid in the small intestine, being denser with increased in the HU in the post-angiographic scans **b** when compared with pre-angiographic CT images **a** indicating a possible communication between the artery, mass (*white arrow*) and small bowel intestine

enteric fistula is even rarer with only one case reported in literature [5]. Pelvic vascular fistula formation is associated with prolonged ureteral stenting, pelvic surgery, and vascular disease. Patients who have undergone radiotherapy treatment for pelvic malignancies show a greater tendency to develop UAFs [6]. In our case, the deceased had several predisposing factors for development of a UAF including pelvic malignancy, previous radiation therapy to the pelvis and prolonged bilateral ureteral stenting. Clinical diagnosis of pelvic vascular fistulas is often difficult as routine radiological examination may not show the fistulous communication [7]. This reflects a systemic problem with clinical CT investigation of abdominal vascular fistulas evidenced by a reported accuracy for diagnosis of aorto-enteric fistula ranging from only 18 [8] to 29 % [9].

The presentation of pelvic vascular fistulas varies considerably from minor microscopic and intermittent hematuria, rectal or vaginal bleeding to massive life threatening hemorrhage. The initial, less significant bleed as reported in this case has been referred to as the "herald bleed" and is felt to be a significant feature of the disease process [10]. Death from massive hemorrhage ranges from 14 to 23 % [11].

Conventional autopsy is usually required for the definitive diagnosis of pelvic vascular fistulas although dissection can

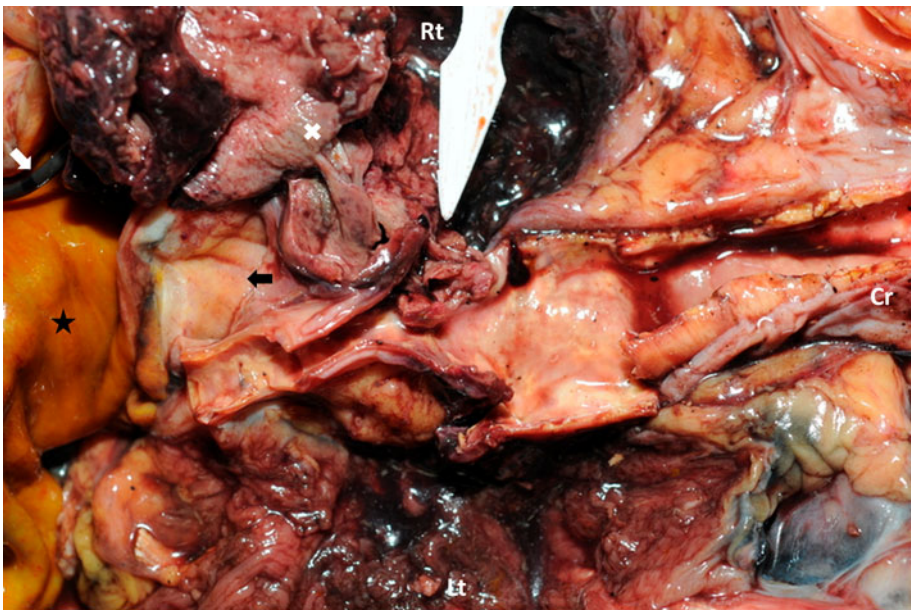

Fig. 4 Autopsy image of the large fibrotic and necrotic mass in the right pelvic wall, which involved the bladder (*black star*), distal right ureter with a ureteric stent in situ (*white arrow*) and a portion of the small intestine (*white arrow*) with erosion through the medial side of the right external iliac artery (*white indicator*). The *cranial side* of the autopsy specimen is indicated by "CR"

be complicated especially in cases of pelvic malignancy treated with surgery and radiation therapy. The tumor mass in these situations is usually necrotic and clumped together with adjacent anatomical structures. It can be challenging for the forensic pathologist to dissect the mass and separate it from surrounding tissues without disrupting the anatomy.

Post-mortem computed tomography (PMCT) scanning of the deceased, even when autopsy is to be performed, is valuable for the forensic pathologist as it provides an overview of the deceased's anatomy and in many cases an indication of likely pathology allowing the pathologist to tailor autopsy technique[12]. It helps to guide dissection [13] especially in anatomical sites that are difficult to access during autopsy, for example the pelvis. In this case, however, PMCT provided only limited diagnostic information for the pathologist. There was no obvious intra-abdominal or pelvic hematoma although a mass was detected in the right lateral pelvic wall.

In clinical practice radiographic contrast would be administered intravenously at the time of CT. This is not routinely performed during PMCT so diagnosis of vascular pathologies is normally limited [14]. Recent literature has shown that PMCTA increases the accuracy of vascular lesion detection [15]. PMCTA is a recently described technique in which PMCT is combined with arterial infusion of radiographic contrast [16–19]. There are many different PMCTA techniques described in literature but there is no single technique appears to be the gold standard. They vary from selective single organ system to whole body angiography. Some centers prefer oil-based contrast solution over water-based contrast solution but each with its own advantages and disadvantages [20]. The decision on either to invest on an embalming machine or a modified pressure-controlled heart–lung machine totally depends on each center's financial budget, dedicated trained staff, number of cases and personal preferences. The balance must be met between developing a cost-effective, minimally invasive, high throughput methodology but avoiding causing angiography related trauma/artifacts or dislodging fresh occlusive thrombus [21].

In our center, we use a modified Ross et al. [16] technique using an embalming pump with a water-based contrast solution and PEG200 in a 1:10 concentration. The embalming machine was preferred over the modified heart–lung machine considering the relatively cheaper price and simpler operation procedures which does not require highly trained staff. However, infusion pressure monitoring is not possible with the embalming machine as compared to the modified heart–lung machine.

The decision to perform PMCTA at our Institute is only entertained following a review of the circumstances of death suggesting hemorrhage, review of PMCT findings and discussion with the forensic pathologist. Given the circumstances of massive external bleeding in this case, presence of a mass lesion on PMCT in close proximity to calcified right iliac vessels and a request for improved information prior to autopsy by the forensic pathologist, PMCTA was undertaken. Contrast leak was clearly seen passing from the right external iliac artery entering the pelvic mass and then into the right ureter. Images also showed the high density contrast fluid in small intestine, being denser in the PMCTA images when compared with PMCT images. These findings were highly suggestive of a communication between the artery, mass, right ureter and small intestine and were confirmed during autopsy. This

clearly explained the clinical presentation of massive hemorrhage from both vagina and rectum.

Conclusion

Ureteric–arterial fistulas (UAF_ remains one of the most difficult cases to diagnose both in clinical and post-mortem settings. PMCT and PMCTA in this case following pelvic surgery and radiotherapy, was an excellent adjunct to conventional autopsy. The final diagnosis of ureteric–arterial–enteric fistula causing massive bleeding as the cause of death was established prior to autopsy and confirmed at autopsy, although confirmation of underlying pathology required histological analysis of tissue obtained at autopsy showing only necrotic post-radiotherapy change rather than tumor. This case exemplifies the supplementary role of post-mortem imaging to the investigation of cause and manner of death in modern medico-legal death investigation.

Key points

1. UAF is difficult to diagnose in both clinical and post-mortem settings.
2. Ureteric–arterial–enteric fistula following pelvic surgery and radiotherapy is extremely rare.
3. PMCTA was a useful adjunct to conventional autopsy in the diagnosis of a lethal UAF.

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Conflict of interest We declare that the authors have no conflicts of interest.

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