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Coexistence of isolated internal iliac artery aneurysm with uterine cervical carcinoma

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Abstract: Isolated internal iliac artery aneurysms are rarely described in the available literature. The paper presents a case of a 70-year-old female with idiopathic thrombocytopenia, squamous cell cervical carcinoma, and saccular aneurysm of the left internal iliac artery, detected in magnetic resonance. The review of aneurysm of the common, external and internal iliac arteries is added.

Keywords: internal iliac artery aneurysms, cervical carcinoma, aneurysm epidemiology, aneurysm risk factors, aneurysm symptoms.

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Introduction

Atherosclerosis, copper deficiency and infections, in particular syphilis, are the main risk factors of aneurysm, which is defined as local dilatation of an artery. It may occur in any blood vessel, but the most common localizations include the aorta, heart, as well as iliac, cerebral and renal arteries [1]. The paper reports a rare case of a saccular aneurysm of the internal iliac artery in a patient with advanced cervical cancer.

Case presentation

A 70-year-old retired female farmer (gravida 6, para 4, with a history of a single previous Cesarean section) was admitted to hospital due to histologically confirmed cervical squamous cell carcinoma (G2). The patient was initially deemed ineligible for surgical treatment due to idiopathic thrombocytopenia (56.000/μl; normal range: 130.000 to 400.000/μl). All evaluated laboratory parameters were initially unaffected, with the exception of slightly longer prothrombin time (12.6 s; normal range: 9.4 to 12.5 s), increased level of cancer antigen 125 (CA125: 44.9 IU/ml, normal range: 0.0 to 30.2 IU/l), as well as slightly decreased level of hemoglobin (11.4 mmol/l; normal range: 12.0 to 16.0 IU/l), and hematocrit (36.1; normal range: 37.0 to 47.0). She suffered from chronic arterial hypertension, well-managed with enalaprili maleas. Cholecystectomy was performed few years previously due to cholecystolithiasis. Besides the above mentioned, medical history was otherwise unremarkable.

Physical examination revealed a solid tumor involving the uterine cervix and distal part of the vagina (stage IIA/IIB according to FIGO), enlargement of inguinal lymph nodes, as well as trophic changes on the skin of the lower limbs. Abdominal sonography and chest X-ray did not show any abnormalities, apart from some atherosclerotic lesions in non-dilated aorta. A routine pelvic magnetic resonance (MR) was performed on 1.5T MR scanner (Achieva; Philips Medical Systems; Veenpluis, The Netherlands) to establish local stage of cancer in accordance with the standard protocol, which included coronal, sagittal and axial series with 4 mm section thickness [2]. The following sequences were performed: T1- and T2-weighted turbo spin echo, with (SPIR – Spectral Presentation with Inversion Recovery) and without fat suppression and WAVE (Water Selective Volume Excitation), diffusion weighted images (DWI; b = 0, 50, 500 and 1000 s/mm^2), followed by ADC map (Apparent Diffusion Coefficient). Additional coronal (dynamic and static) and sagittal T1-weighted images (static) were obtained after gadolinium-diethylene-triamine-penta-acetic acid (Gd-DTPA) injection (0.1 mg/kg). Data were stored on Picture Archiving and Communication System (PACS) and examined on Extended MR Work Space 2.6.3.4. (Philips Medical Systems; Veenpluis, The Netherlands).

The examination revealed advanced tumor of the cervix (39 × 33 × 35 mm, AP × W × CC; v = 22 ml), with irregular border and contrast enhancement, as well as partially calcified intramural myoma of the uterine body (Fig. 1). Infiltration of the parametrium/paracervix and the upper 1/3 of the vagina was observed (Fig. 2). Infiltration of the urinary bladder and rectum was excluded. Mean value of ADC was 0.99×10^{-3} mm^2/s ($0.69–13.2 \times 10^{-3}$ mm^2/s). Bilaterally, 6 parametrial lymph nodes with cross diameter up to 8 mm but with high ADC values ($>1.2 \times 10^{-3}$ mm^2/s) were observed. Single, borderline left external (10 mm) and bilateral enlargement of the superficial inguinal lymph nodes were found (up to 21 mm). All of the remaining

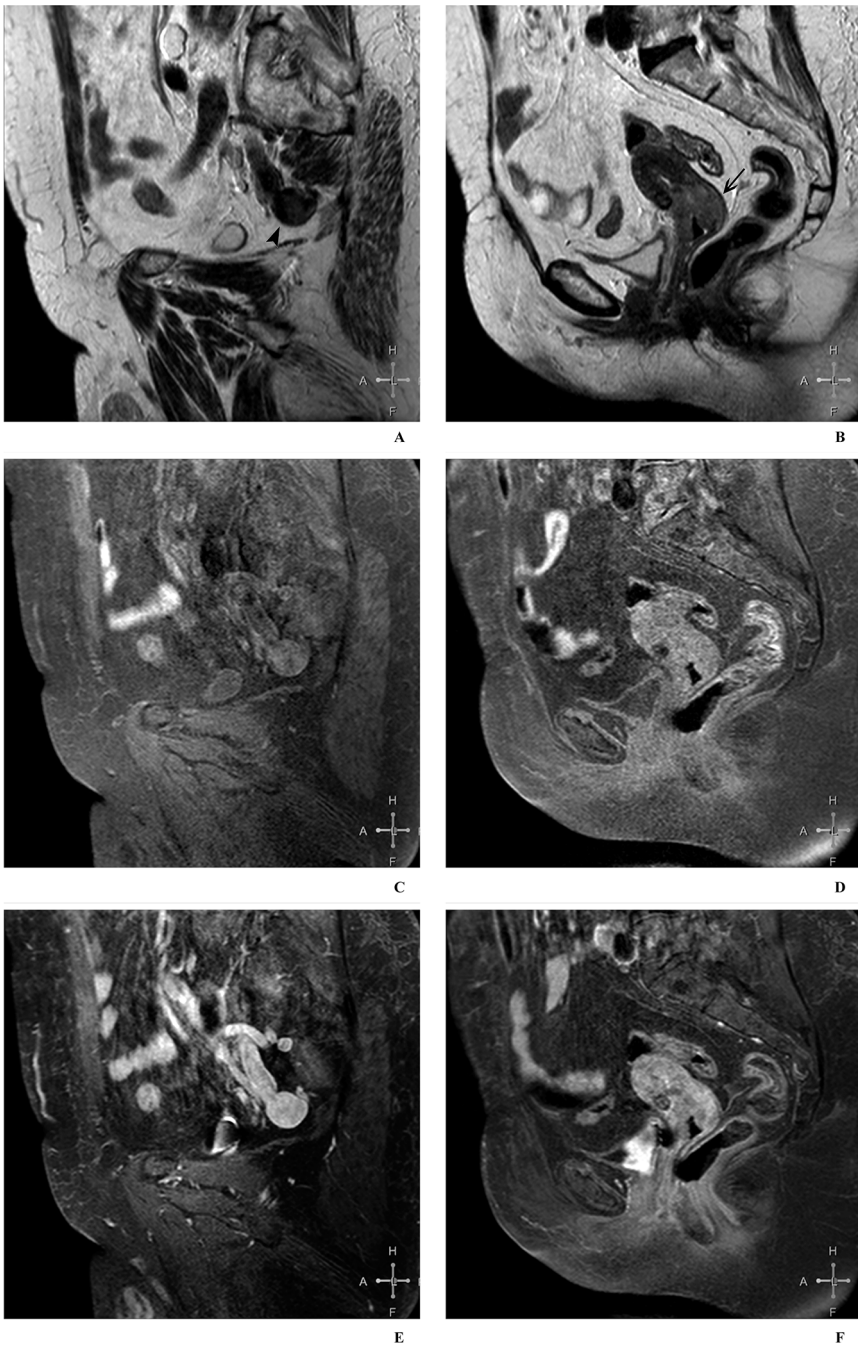

Fig. 1. The saccular aneurysm of the terminal part of the left internal iliac artery (arrow head) and cervical cancer (arrow) on sagittal section. A–B — T2-weighted image, C–D — T1-weighted image with fat saturation, E–F — T1-weighted image with fat saturation and contrast enhancement (late phase).

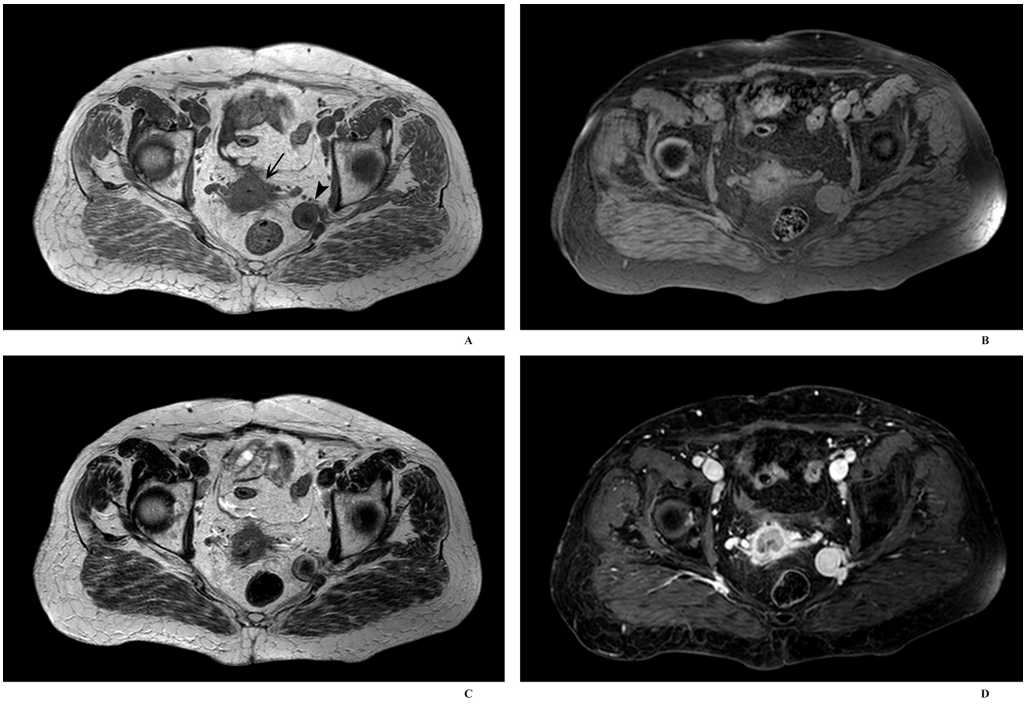

Fig. 2. The saccular aneurysm of the terminal part of the left internal iliac artery (arrow head) and cervical cancer (arrow) on axial sections. A — T1-weighted image, B — WAVE image, C — T2-weighted image, D — T1-weighted image with fat saturation and contrast enhancement (late phase).

nodes were small, reaching the cross diameter up to 6 mm for the external iliac, as well as 7 mm for internal and common iliac nodes.

Inside the pelvis, on the level of the left ischial spine a local, saccular enlargement (25 × 23 × 26 mm) of the left internal iliac artery was found (Fig. 1 and 2). Slow contrast enhancement was seen during dynamic examination (Fig. 3). Furthermore, low volume of peritoneal fluid as well as degenerative vertebral changes and intervertebral disc protrusion (L5–S1) were revealed.

The final diagnosis confirmed cervical carcinoma (stage IIB, T2N1Mx) with concomitant saccular aneurysm of the left internal iliac artery and degenerative vertebral changes. The patient was referred to radiotherapy and a vascular consultation was advised during multi-disciplinary team meeting.

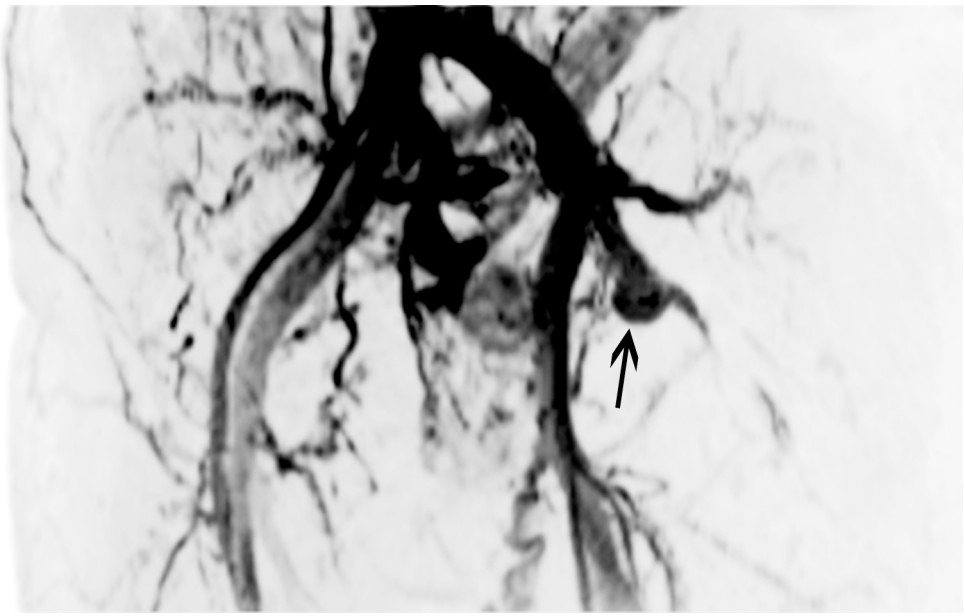

Fig. 3. The saccular aneurysm of the terminal part of the left internal iliac artery (arrow head) on pseudo-3D vascular reconstruction after contrast injection (positive).

Discussion

The presented localization of the aneurysm is extremely rare and seldom reported in the available literature. The incidence of isolated internal iliac artery aneurysms is less than 2% of all abdominal aneurysmal diseases [3]. More commonly they coexist with aortic aneurysms (10–20%). Similar low occurrence was reported in a historic autopsy study, where 321 aneurysms (including 3 discussed) were found among 12.000 bodies [4]. On the other hand, Dix *et al.* [5], in their search of PubMed, Medline and Medscap database, were able to find only 94 atherosclerotic aneurysms in such localization. Median age of all patients was 71.9 (47–89) years, with corresponding increased risk of rupture, which was revealed in 40% of the cases, leading to rapid death if untreated. Most pathologies (95%) were observed among male patients, with median (range) size of 7.7 (2–13) cm. Higher male-female ratio (6:1) and lower mean age (67.2 year) were reported previously by Brin and Busuttil [6]. Younger patients were also seen, especially among females who were more commonly diagnosed during pregnancy. Based on their data, arterial wall changes, mostly of non-specific nature, were found as the main risk factor in 80% of the cases. Traumatic etiology was pointed out in 11% of the affected individuals, particularly among females reporting traumatic childbirth, high forceps deliveries, or cesarean section. Similar to aortic aneurysm, local bacterial

infections have been also stressed, including *Staphylococcus aureus*, *Klebsiella spp*., *Pseudomonas spp*. and *Salmonella spp*. [7]. Higher risk was also reported for fibromuscular dysplasia, cystic medial necrosis, and collagen vascular diseases [1]. At present, the incidence of the discussed aneurysm will probably increase since various authors report higher occurrence in patients previously surgically treated for aortic and/or common iliac aneurysm [1, 3, 5]. Progressive expansion is natural for isolated iliac artery aneurysm, but its rate significantly depends on the size [8]. Lesions smaller than 3 cm expand at an average rate of 0.05–0.15 cm/year, while larger than 3 cm increase at up to 0.28 cm/year.

Symptoms of isolated internal iliac artery aneurysms are usually non-specific and related to compression of adjacent structures, local expansion, fistulation and rupture [7]. They usually include abdominal pain (31.7%), urological symptoms (28.3%), groin pain (11.7%), hip or buttock pain (8.3%), and other neurological (18.3%) as well as gastrointestinal symptoms (8.3%) [5]. Most symptoms are related to compression of the surrounding structures, especially ureter and bladder, observed in 15–45% of the cases, that usually case difficult and pulsatile micturition, hematuria, hydrouretronephrosis and higher risk of urinary infections. Less commonly, the lumbo-sacral plexus and its nerves are compressed (10–15%), leading to sciatic, femoral, pudendal and/or obturator neuropathies (sacral neuralgia, hypoaesthesia, paralysis, loss of sphincter tone). The plexus may also be affected by ischemic changes due to impression and thrombosis of its own nutrient arteries. Moreover, vascular complications including venal insufficiency and acute ilio-femoral thrombosis have been seen in 5% of patients. Occasionally, especially in big aneurysm, constipation due to the compression of the rectum is observed. More commonly, symptoms related to leakage have been seen. Similar to aneurysm, the pelvic hematoma may induce compressive symptoms. Extra-peritoneal rupture that usually enters rectum or bladder, but isolated perianal and valvular ecchymoses were also described. However, Brin and Busuttil stress that 43% of the vasculopathy was asymptomatic and incidentally detected [6]. Moreover, Richardson and Greenfield postulated that 55% of the affected patients have a pulsatile mass, palpated easily during the digital rectal and/or transvaginal examination [7]. Lower incidence (20%) of such symptoms was reported by Krupski *et al.* [9].

Proper and fast diagnosis is vital since aneurysmal rupture at the time of diagnosis has been reported in 33–38% of the affected individuals [6, 10]. Furthermore, mortality reaches 58%, and is positively associated with lesion size [6, 7]. The diagnosis should be easy, since most aneurysms can be visualized using ultrasound, computed tomography, MR or angiography, which may be performed directly before endovascular treatment [1, 3, 7–12]. Surgical treatment is challenging but achievable by ligation, excision or endoaneurysmorrhaphy. However, interventional radiological procedures, including coil embolization and endoluminal stenting (alone or in combination, have been more commonly applied as the alternative, since such treatment is

feasible and safer, with lower morbidity and mortality rates [5]. However, on average more than one intervention has to be performed to achieve successful permanent exclusion of the aneurysm. Embolization alone in isolated internal iliac artery aneurysms is not sufficient [12].

Coexistence of the iliac artery aneurysm and gynecologic malignancies is rare and confined to few cases described in the available literature. However, up until now, such complication was always reported for ruptured aneurysms, usually limited to the common and external iliac artery [13–15]. In eight cases, previous surgery and external beam radiotherapy (mean 30–65 Gy, 3 months to 20 years before hemorrhage) had been performed, including six cases of advanced cervical cancer (3 treated with an additional cervical brachytherapy — 10–25 Gy) [14, 15]. Among seven women reported by de Baere *et al.*, in two cases bleeding was limited to the aneurysm of the internal iliac artery, but precise cancer localization was not presented since the whole group included seven females and one male: cervical carcinoma (n = 5), rectal adenocarcinoma (n = 2), and iliac bone fibrosarcoma (n = 1) [15]. The remaining ones were limited to the external iliac artery (n = 2), common iliac artery (n = 2) and its bifurcation (n = 1). Additionally, the above presented case of cervical squamous cell carcinoma reports bleeding from the external iliac artery [14]. Unlike gynecological malignancies, more commonly the coexistence was reported for rectal neoplasms as well as aortic aneurysm with different abdominal and thoracic malignancies. Higher incidence of aneurysms among patients treated with radiotherapy could be explained by ionizing-related changes of the small arteries that supply the wall of the bigger vessel, with secondary degenerative changes that led to their local dilatation or narrowing [15]. However, atherosclerotic lesions and vascular thrombosis remain the most frequently reported vascular pathologies in oncological patients [16].

Conclusions

To the best of our knowledge, this has been the first report on unruptured aneurysm of the internal iliac artery, concomitant with cervical cancer. Due to patient obstetric history (gravida 6, para 4) it is probably a pseudo-aneurysm. However, based on radiological data, it is not possible to state the etiology of the lesion, especially since idiopathic thrombocytopenia was previously diagnosed in the patient. Such vascular lesions are rare, often asymptomatic and usually incidentally observed, as was the case in the presented patient.

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