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Misinterpretation of a giant idiopathic concealed right iliac artery aneurysm as infantile pelvic tumor imaged by non-angiographic computed tomography: a case report

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Abstract

Background Idiopathic pediatric artery aneurysm is extremely rare and sometimes behaves like a pelvic neoplasm. As it is associated with a high mortality while ruptured, the accurate diagnosis is indispensable to avoid life-threatening complications. Angiographic imaging is the gold standard for diagnosis and treatment planning of the aneurysm. Due to the considerably higher radiation sensitivity of children, single portal venous-phase scanning of the abdominopelvic computed tomography (CT) is, however, reasonably utilized for the evaluation of a clinically palpable mass; the erroneous diagnosis of such aneurysm can be feasible.

Case presentation A giant idiopathic concealed right iliac artery aneurysm in a 47-day-old girl presented with a palpable pelvic mass regardless of unidentifiable predisposing factors. Non-angiographic abdominopelvic CT was reappraised by the radiology consultant according to discordance between the mass characteristics on initial CT report and those on second-look sonography, revealing the concealed aneurysm instead of solid neoplasm as it originated from right internal iliac artery. The patient underwent an emergency laparotomy with successful proximal ligation of right internal iliac artery despite intraoperative aneurysmal rupture.

Conclusions Typical CT features for the infantile iliac artery aneurysms may be overlooked, especially if the angiographic phase is omitted; thus, the imaging characteristics of the aneurysms are more difficult to appreciate and can mimic a pelvic neoplasm. Therefore, the identification of the origin of the mass should be more practical to achieve the precise diagnosis.

Keywords Idiopathic aneurysm, Iliac aneurysm, Congenital, Computed tomography, Case report

Background

Idiopathic or congenital pediatric iliac artery aneurysm is extremely rare, and fewer than 20 reported cases have been published until 2021 in English-language literature [1–6]. The etiology is unknown [7]. Despite the rarity of occurrence, early recognition of the aneurysm is important to avoid the potentially fatal complications, such as ruptured aneurysm and renal failure [7, 8]. Generally, sonography is an initial imaging for children presenting with lump abdomen. Computed tomography (CT) or magnetic resonance angiography is recommended as noninvasive modality of choice for assessment and treatment planning of the vascular mass [8]. In routine practice, especially if sonography is unavailable, the single portal venous-phase CT is often applied to evaluate clinical abdominopelvic mass with the

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diagnostic accuracy of 100% [9, 10]. Herein, we described an infant with palpable abdominal mass where a single portal-venous-enhanced CT study demonstrated a large enhancing pelvic mass concerning for gynecologic neoplasm. However, because of lacking consistency with sonographic findings, the definite diagnosis was revised to be a concealed iliac artery aneurysm confirmed by the subsequent operation.

Case presentation

A 47-day-old female infant was transferred to our tertiary referral hospital, presenting with a palpable non-pulsatile abdominal mass for 10 days. Three days before the referral, she then developed swelling and purplish discoloration in her right groin and concurrently progressive oliguria while staying in the general hospital. Two days later, abdominal CT included unenhanced, and portal venous-phase images were interpreted as a large pelvic neoplasm, e.g., sarcomas. Apart from that, the girl born in the community hospital at 39-week gestation by uncomplicated vaginal delivery was healthy with Apgar scores of 9/10 and a birth weight of 3430 g. Her mother, a 27-year-old Thai multigravida (para 2, gravida 3, abortion 0, live children 2), was in denial of underlying disease, familial health problem, and medication. The antenatal and postnatal records were unremarkable.

Due to the diagnosis of pediatric pelvic tumor based on the radiology report, the patient was admitted to our non-intensive care ward. Her vital signs were stable. On physical examination, a large, firm, non-pulsatile, fixed mass was palpable in suprapubic region along with dilated superficial abdominal veins, as well as swollen right leg and purplish discoloration in the bulging right groin (Fig. 1). There was no diminished lower-extremity peripheral pulse. The remainder of the examination was unremarkable. Initial basic laboratory tests revealed no abnormality other than severe anemia with a hemoglobin level of 6.5 g/dL and a hematocrit level of 19%. Many serum tumor markers measured for possible gynecologic tumor pathologies were normal. A bedside ultrasound for further evaluation regarding oliguria demonstrated a huge cystic mass in the pelvic cavity which was apparently discordant with the recent CT radiology report. Thus, second-opinion radiology review of the abdominopelvic CT proposed a large lobulated enhancing mass of $7.9 \times 7.4 \times 6.5$ cm in right side of the pelvic extraperitoneal compartment, iliopsoas muscle, and inguinal region arising from right internal iliac artery, together with a crescentic, eccentric, non-enhancing, hyperattenuating collection along the posterior itself (Fig. 2). It was thought to be a giant concealed right internal iliac artery aneurysm with retroperitoneal hematoma. Abdominal aorta and the remainder of iliac arteries were patent. An emergency laparotomy was performed immediately when the patient received sufficient fluid resuscitation and blood transfusion. We discovered the concealed aneurysm. Right internal iliac artery proximal to the aneurysm was promptly ligated together with massive blood transfusion and cardiopulmonary resuscitation owing to intraoperative rupture of such aneurysm with active bleeding and profound hemodynamic instability. The patient then underwent re-exploration and percutaneous nephrostomy in consequence of jejunal perforation and right ureteric injury. On the 1-month postoperative CT, the aneurysm was thrombosed. Thereafter, she was fully recovered and discharged on the 72nd day of hospitalization. The infant was followed up in the

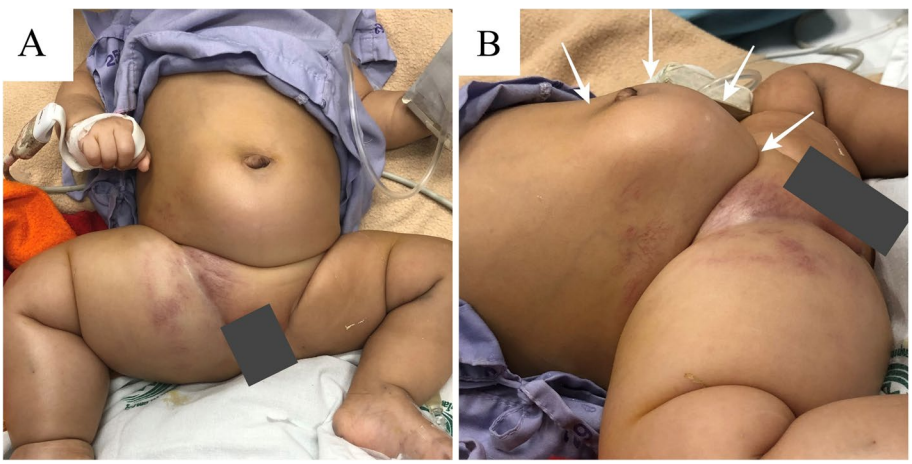

Fig. 1 Focused pictures at lower abdomen and thighs in frontal (**A**) and lateral (**B**) approaches. There was focal bulge (white arrows) with purplish skin discoloration at right side of lower abdomen and right groin. Asymmetric swollen right leg was also seen (**A**–**B**)

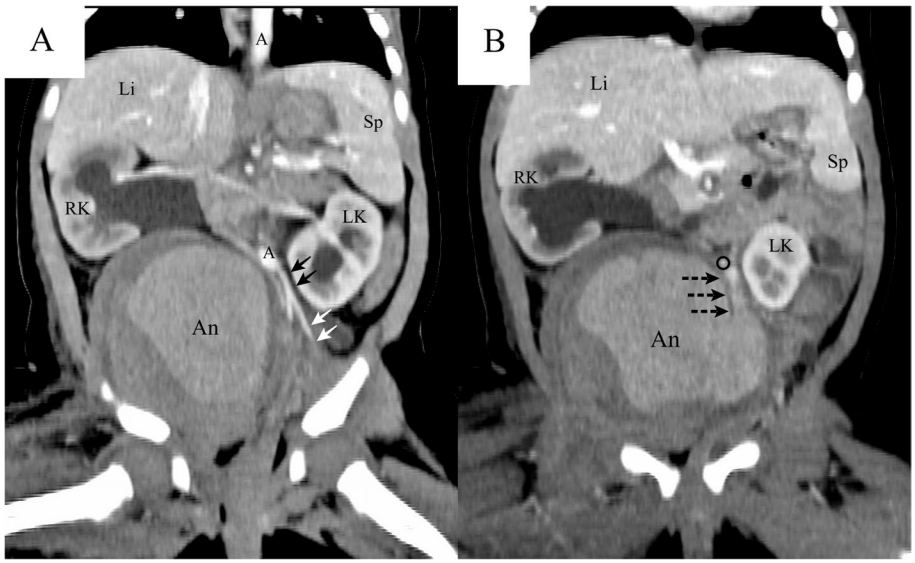

Fig. 2 Coronal non-angiographic CT images at level of aortic bifurcation (**A**) and of right iliac artery bifurcation (**B**). There was a giant right internal iliac artery aneurysm with surrounded hematoma in right side of pelvic cavity (**A**–**B**). The abdominal aorta, left common iliac artery (black arrows), and left external iliac artery (white arrows) were not involved (**A**). The aneurysm arose from right internal iliac artery (black dash arrows) with patency of right external iliac artery (black circle) (**B**). Bilateral hydronephroses were depicted. Abbreviations: A, aorta; An, aneurysm; Li, liver; LK, left kidney; RK, right kidney; Sp, spleen

pediatric and urology outpatient clinics without any other complications observed.

Discussion

Rising radiation awareness from the pediatric CT imaging may have an influence upon radiologic decisions to design individual pediatric technical protocol for radiation reduction. In general, a single portal venous phase is practical and sufficient for the mass determination in the children with a clinical suspicion of abdominal neoplasms [9]. Recently, Gupta et al. proposed the accuracy of the single-phase CT in terms of the detection, localization, determination of the extension, and the diagnosis in solid pediatric abdominal tumors was quite high (81–100%) [10]. Yet, the diagnostic performance in other diseases particularly congenital aneurysms is still unknown.

Angiographic phase is the gold standard technique for assessment of the aneurysms. It is performed during peak aortic enhancement to ensure maximum arterial opacification and reduce venous contamination that provides notably vascular visualization [8]. Meanwhile, the portal-venous phase is performed during maximum hepatic parenchymal enhancement where the attenuation of solid organs or tumors may be virtually indistinguishable from the vascular pathologies. Therefore, CT features suggestive of aneurysms in non-angiographic CT are not widely available due to unstandardized practice. In our experience, searching organ of origin by identifying anatomical relationship with relevant adjacent structures should be more beneficial for the precise diagnosis of uncommon aneurysms despite lack of angiographic phase.

The congenital pediatric aneurysm is an extremely rare disease entity, especially when it occurs at iliac arteries. Clinical presentation might range from asymptomatic to rupture [7]. Among the literature review, the most common includes a palpable abdominal mass or lack of symptom, while the far less common is a rupture [1–8]. The relevant imaging, e.g., ultrasonography, CT or magnetic resonance angiography, and digital subtraction angiography, is necessary for diagnosis and treatment planning [8, 10]. The previously reported cases almost started imaging investigations with ultrasound for initial evaluation of the aneurysms, followed by CT angiography except that one reported only used non-angiographic CT-proven, right iliac artery aneurysm [1–4, 7].

In our patient, the single portal venous-phase CT was performed with a clinical indication of pediatric pelvic mass that the significant radiologic misinterpretation occurred. However, it was fortunate that ultrasound re-evaluation helped make diagnostic discordance. Second-opinion radiology review defined it as a concealed right iliac aneurysm owing to determination of the vascular origin. Therefore, we realized that the radiologic error might be because of the power of persuasion by a clinical suspicion of the neoplasms,

lack of angiographic phase, disease rarity, and an inexperienced radiologist.

Conclusion

Non-angiographic abdominopelvic CT for the infantile pelvic masses is challenging to differentiate congenital iliac artery aneurysms from the much commoner pelvic neoplasms because typical features for the aneurysms may be missed and enhancement pattern is more difficult to appreciate. As the imaging characteristics of two disease entities may be overlapped, the identification of the nature of masses by using anatomical relationship with relevant adjacent structures should be more practical to achieve the right diagnosis.

Abbreviations

CT Computed tomography

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Authors' contributions

TT and TT contributed to the case conception and collected the data. TT wrote the first draft of the manuscript. The authors read and approved the final manuscript.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on request.

Declarations

Ethics approval and consent to participate

Exemption from ethical approval was obtained from the Institutional Review Board of Buddhachinaraj Hospital Phitsanulok (IRB number 015/65). Verbal informed consent was obtained from the mother to participate in the case report and prior to taking photographs.

Consent for publication

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

Competing interests

The authors declare that they have no competing interests.

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References

1. Lee JH, Oh C, Youn JK, Han JW, Kim HY, Jung SE. Right iliac arterial aneurysm in a 4-year-old girl who does not have a right external iliac artery. Ann Surg Treat Res. 2016;91:265–8.
2. Krysiak R, Żyłkowski J, Jaworski M, Brzewski M, Bober-Olesińska K. Neonatal idiopathic aneurysm of the common iliac artery. J Vasc Surg Cases Innov Tech. 2019;5:75–7.
3. Chithra R, Sundar RA, Velladuraichi B, Sritharan N, Amalorpavanathan J, Vidyasagaran T. Pediatric isolated bilateral iliac aneurysm. J Vasc Surg. 2013;58:215–6.
4. Lopez-Gutierrez JC, Rodriguez LC, Zurita MB, Contreras CU, Ákvarez-Luque A, Prieto C. Multiple congenital ectatic and fusiform arterial aneurysms associated with lower limb hypoplasia. J Vasc Surg. 2012;56:496–9.
5. Hoshiko FM, Zampieri EHS, Dalio MB, Dezotti NRA, Joviliano EE. Repair of ruptured iliac artery aneurysm in a child. J Vasc Bras. 2017;16:48–51.
6. Davis FM, Eliason JL, Ganesh SK, Blatt NB, Stanley JC, Coleman DM. Pediatric nonaortic arterial aneurysms. J Vasc Surg. 2016;63:466–76.
7. Wang Y, Tao Y. Diagnosis and treatment of congenital abdominal aortic aneurysm: a systematic review of reported cases. Orphanet J Rare Dis. 2015;10:1–7.
8. Restrepo R, Ranson M, Chait PG, Connolly BL, Temple MJ, Amaral J, et al. Extracranial aneurysms in children: practical classification and correlative imaging. AJR. 2003;181:867–78.
9. Petit S, Vallin C, Morel B, Bertrand P, Blouin P, Lardy H, et al. A single-enhanced phase is sufficient for the initial computed tomography evaluation of retroperitoneal tumors in children. Diagn Interv Imaging. 2017;98:73–8.
10. Lucena IRS, Chedid MF, Isolan PS, Takamatu EE, Lucena RA, Feier FH, et al. A comparison between ultrasonography and single-phase computed tomography for preoperative assessment of solid abdominal tumors in children. Jornal de Pediatria. 2023;99:17–22.

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