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CASE REPORT

Co-existing Isolated Internal Iliac Artery Aneurysm and Solitary Kidney: A Cadaveric Case Study

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Abstract

Purpose: The purpose of this case study is to report on an isolated internal iliac artery aneurysm with a solitary kidney found during cadaveric dissection and to discuss potential clinical implications of such a variation.

Case Presentation: An isolated internal iliac artery aneurysm and solitary kidney were discovered during cadaveric dissection performed on a male cadaver by six nursing graduate students at Samford University in Birmingham, Alabama. Upon discovery, a literature review was performed to consider clinical implications, patient presentation, and management strategies associated with an isolated internal iliac artery aneurysm and solitary kidney. During the literature review, no other documented cases of the pairing of these variations were found. It is unknown whether or not this person had symptoms associated with either of these conditions.

Conclusion: As these variations were discovered post-mortem, it is unknown whether this condition was symptomatic.

Key Words: *Internal iliac artery; Aneurysm; Solitary kidney; Isolated internal iliac artery aneurysm, Renal anomalies*

Introduction

The following case study elaborates upon two rare anatomic variations found during cadaveric dissection on a male cadaver in a graduate nursing anatomy course. Isolated internal iliac artery aneurysm (IIIAA) is extremely rare [1-4]. A solitary kidney is another rare condition [5]. These two phenomena, on their own, are rare in an individual; the two of them together further reveal the uniqueness of this case. While it is documented that a solitary kidney can lead to hypertension [6], a risk factor for an internal iliac artery aneurysm (IIAA) [7], this appears to be the first reported case of these two anatomical variations present together. The case report reported here did not require Institute Review Board review or approval; all students signed a Pledge of Respect to the person donating his remains for educational purposes.

Case Presentation

Students discovered the cadaver possessed an isolated internal iliac artery aneurysm (IIIAA).

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Findings on dissection measured the IIIAA as approximately 5.3 cm in circumference and 2.5 cm in length. Figure 1 shows the IIIAA found during cadaveric dissection. Additionally, the cadaver had a solitary kidney - not possessing a left kidney or left renal artery (Figure 2).

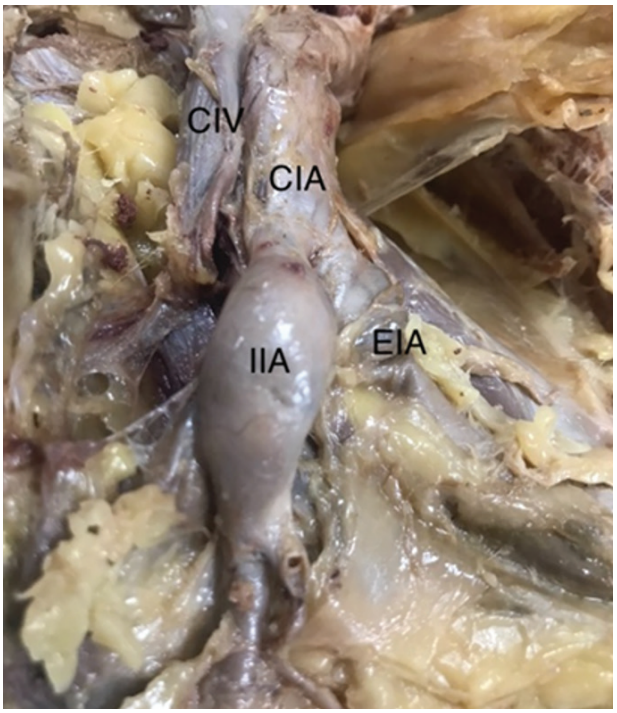

Figure 1) *Common Iliac Vein (CIV), Common Iliac Artery (CIA), External Iliac Artery (EIA), and Internal Iliac Artery (IIA).*

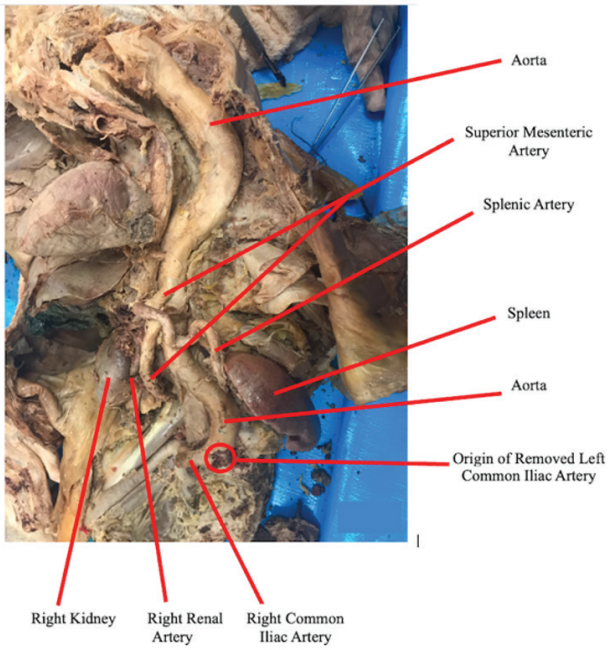

Figure 2) *Solitary right kidney prior to removal of vascular tree.*

Discussion

IIIAA

Incidence and prevalence

An isolated internal iliac artery aneurysm is defined as the presence of an IIIAA without simultaneous representation of an aneurysm in the abdominal aorta, common iliac artery, or external iliac artery. Reports of an IIIAA occur at a prevalence of 0.008-0.03% according to case reports and autopsy series, representing 0.3-0.5% of all intra-abdominal aneurysms [1-4,8-12]. Of IIIAAs reported, most patients diagnosed are 65-84 years old with a median aneurysm diameter of 6.0 cm [1-4,11,12]. The presence of IIIAA is reported in a 6:1 male-to-female ratio, or 86-92.7% male [1-4,11,12]. The most common etiology leading to the pathogenesis of IIIAAs is related to progressive arteriosclerosis, infection, trauma, connective tissue disorders, and cesarean section delivery [2,4]. Interestingly, a left-sided IIIAA appears more than twice as often as a right-sided IIIAA at a rate of 61.8% and 27.3%, respectively [12]. The remaining 10.9% are bilateral [12].

Patient presentation

The most common clinical findings associated with an IIIAA are palpable masses on abdominal or rectal exams performed to assist in the diagnosis of patients presenting with abdominal pain, urinary habit changes, or hydronephrosis [12]. Patients may also experience neurological, local, anterior, or posterior thigh, hip, testicular, or lower back pain [9,12]. If an IIIAA is palpable via abdominal or rectal exam, the average size of the aneurysm at this time is reported to be 9.6 cm, which is associated with an increased risk of rupture [12]. Of palpable IIIAA masses reported, 32.7% are found with abdominal palpation, and 21.8% by rectal examination [12]. Bleeding associated with a ruptured or leaking IIIAA will usually present

via the genitourinary or gastrointestinal systems [9]. Frequently reported neighboring structures to the IIA impacted by compression from the growing IIIAA include: (a) bladder and ureters, (b) rectum, (c) sciatic nerve, and (d) iliac veins [1,2,4,7-14].

Bladder and ureter involvement with IIIAA

Urologic symptoms are commonly reported in association with an IIIAA at a rate of 40-54% of cases [4]. Genitourinary symptoms of IIIAA are directly attributable to the compression of bladder and ureteral structures and commonly present as: (*a*) hydronephrosis, (*b*) pyelonephritis, (*c*) hematuria, (*d*) dysuria, (*e*) nocturia, (*f*) pulsatile urinary stream, (*g*) renal colic, and even (*h*) renal failure [7,9,12]. There has been a report that 16% of patients with an IIIAA have experienced genitourinary obstruction [9]. The high frequency of ureter involvement is attributed to its susceptibility to obstruction by IIIAA, frequently producing urinary tract infections, bacteremia, and hematuria [15].

Rectal involvement with IIIAA

While constipation as the sole symptom of IIIAA is rare, there have been reports of patients suffering from constipation for up to 2.5 months as a result of an IIAA measuring from 5.6 cm to 6.9 x 6.7 cm [8,9]. These symptoms caused by IIAAs often occur in men aged 47-85 years [9]. Alleviation of constipation attributed to IIIAA occurs after surgical correction; however, in some cases, gastrointestinal involvement from IIIAAs can lead to intestinal ischemia requiring surgical resection and colostomy [11,12].

Sciatic nerve involvement with IIIAA

Compression of the sciatic nerve has been reported in 10-15% of IIIAA cases [4]. Neurologic symptoms present once the IIIAA is sizeable enough to compress pelvic nerves, including the L5 and S1 lumbosacral roots, which are found directly posterior to the IIA. Presentation of symptoms due to compression has been reported as pain radiating from the left buttock to the posterolateral aspect of the left calf and lateral foot, causing weakness in toe and ankle dorsiflexion, which can impair ambulation.

Iliac vein involvement with IIIAA

The situation that involves the IIIAA compressing the iliac vein has been given the nomenclature May-Thurner Syndrome (MTS) [7,13]. This phenomenon has been traditionally known as iliac vein compression syndrome (IVCS) [13]. The etiology of MTS is thought to be due to an acquired lesion or congenital abnormality. The diagnosis is confirmed by Doppler ultrasound, iliolumbar plain computed tomography (CT), deep vein CT, venography, and angiography [13]. Presentation of MTS includes lower limb swelling and pain that may occur over 1 day to 6 months [13]. These symptoms are eerily similar to those of a deep vein thrombosis, which has a high prevalence of occurring along with pulmonary embolism from the IIIAA's direct compression on the iliac vein [13].

Mortality

Discovery of an IIIAA usually occurs during assessment for known multiple aneurysmal diseases or long-term surveillance of a patient that has received abdominal aortic aneurysm repair (AAA) via CT [11]. The spontaneous discovery of an asymptomatic IIIAA on CT occurs in 16.4% of patients, with the average size of the aneurysm being 5.1 cm [12]. There are no reported deaths in this group of individuals [12]. Patients presenting with symptoms from an IIIAA comprise 52.7% of the discovered population with an average aneurysm size of 7.6 cm [12]. Patients possessing a symptomatic IIIAA experience a 10.3% mortality rate if not treated surgically [12]. Of the cases reported,

30.9-60% of patients discover that they possess an IIIAA after it has ruptured [9,12]. The average size of an IIIAA or IIAA at the time of rupture is 8.3 cm, resulting in a 35-58% mortality rate [1,8-10,12]. Mortality from an IIIAA is attributed to its retroperitoneal or intraperitoneal location, deep in the pelvic cavity, resulting in subtle signs of the aneurysm delaying diagnosis, which increases the incidence of spontaneous rupture and hemorrhage. This delay and rupture further exacerbate difficulties with intra-operative management [1,4,6]. The anatomical position of an IIIAA is the most significant contributing factor to mortality [12,15,16].

The anterior and posterior divisions of the internal iliac artery (IIA) usually arise deep within the pelvis, making it difficult for smaller asymptomatic IIIAAs to be detected [12,16]. An undiscovered IIIAA creates synergistic adverse sequelae as it expands unrecognized deep in the pelvic space [12,15]. When symptoms occur due to the presentation of an IIIAA, it is often large enough to either rupture or force physical pressure on neighboring anatomical structures, disrupting normal physiologic function [12,15].

Surgical Repair and Management

Due to the rarity and the common asymptomatic state of IIIAA, the diagnosis of these aneurysms is typically not discovered until they have grown to an alarming size or ruptured. The aneurysm can be detected using computed tomography angiography (CTA), ultrasound (US), and magnetic resonance imaging (MRI) [10-12]. Helical computed angiotomography is the gold standard for diagnosing IIIAA [1]. Recent studies have suggested that elective repair should be considered once the asymptomatic patient's aneurysm measures 30-40 mm or 3-4 cm [2,3,8,12,15]. If discovered with subsequent imaging, the success rate for elective repair for asymptomatic IIAA patients is nearly 99%, with a 0-7% mortality rate [2,10].

The first surgeon to describe and operate on an IIIAA with an open surgical technique was Archibald MacLaren over 100 years ago in 1913 [11,12]. Since then, there have been only 52 reported cases of IIIAA as of 2014 [12]. It has been found that mortality rates with an open surgical approach are high due to considerable blood loss from pelvic dissection [2,16]. Current practice dictates that the open approach is used in emergency situations or when the IIIAA compresses neighboring structures, causing life-threatening signs and symptoms [2]. Coincidentally, one complication of the open surgical technique is damage to these nearby structures, such as the ureter [11]. The major disadvantage of open surgery is the high recurrence rate, as IIA can be refilled retrogradely by its branches. Due to this phenomenon, recurrence rates from open surgery are 33% [11,12]. Endoaneurysmorrhaphy has been recommended to reduce the incidence of retrograde filling of the aneurysm [12]. Due to several major disadvantages to open surgery, it is mainly indicated if the endovascular repair fails, if visceral revascularization is required, or if symptomatic compression of the ureter is involved [11]. Safer surgical interventions for IIIAA have been researched and are recommended options to open repair when possible: (*a*) excision and exclusion, (*b*) proximal and distal ligation, (*c*) endoaneurysmorrhaphy, and (*d*) endovascular stent-graft [2,11,12,16].

Proximal and distal ligation using an endovascular stent graft is the best approach for repairing IIIAA [11,16]. The endovascular approach has long been used for poor surgical candidates as it provides positive results in managing an IIAA that are associated with lower morbidity and a 1% mortality rate [2,3]. In the case of an IIIAA, the ipsilateral common femoral artery approach is effective due to the ability to maintain collateral circulation while the injured site is repaired [16]. It is because of

the distinct advantage of maintaining circulation that the endovascular stent-graft has been used effectively for thoracic and abdominal aortic aneurysms, pseudoaneurysms, occlusive disease, and arteriovenous fistulas as well as IIIAAs [16].

After embolization, the most significant threat to this post-operative patient population is the potential to develop venous thrombosis [8]. Recommendations for patient management after surgical repair of the IIIAA consist of CT imaging on the second post-operative day, again after one, and six months post-operatively [3,16]. There are also reports of follow-up CT imaging performed one year after endovascular repair [16].

Solitary Kidney

Background

In typical renal anatomy, the kidneys are retroperitoneal located between the transverse processes of T12 to L3. The right and left kidneys are posterior to the ascending and descending colon, respectively. The right and left renal arteries branch off the abdominal aorta inferior to the superior mesenteric artery, entering the kidneys at the hilum to supply them with oxygenated blood. The right and left renal veins exit at the hilum and join the inferior vena cava. [17] (Figure 3).

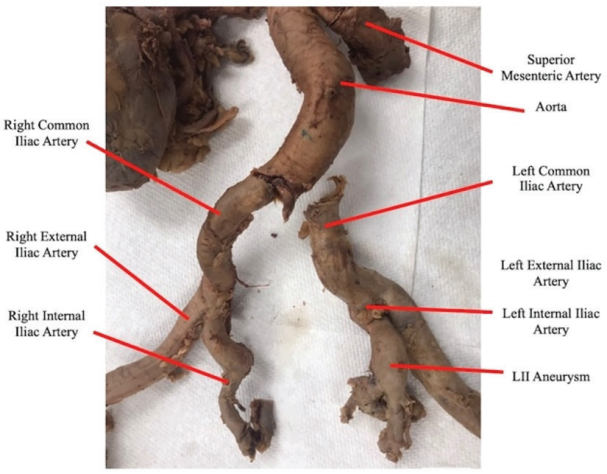

Figure 3) *Vascular Tree with Left Iliac Artery Aneurysm.*

A solitary kidney is characterized by having only one functioning kidney. Various studies have found that those living with one kidney can lead relatively normal lives. When compared to counterparts of similar demographics having two kidneys, comparable risks for cardiovascular disease and mortality were shared. Living with one kidney can cause other comorbidities like hypertension and end-stage renal disease [6]. Renal agenesis, surgical removal, and kidney donation are the three leading causes of a solitary kidney diagnosis. Renal agenesis affects 1 in 2,000 infants globally each year, while kidney donation and nephrectomies total a little over 300,000 Americans [5].

Embryologic development of kidneys involves multiple processes. The metanephric kidney starts with the mesonephric duct fusing with the cloaca, which triggers the sacral mesoderm to form the ureteric bud. The development continues with the ureteric bud leading to creation of collecting tubules, renal lobes, and ultimately arterioles and nephrons. Congenital absence of the kidney, or renal agenesis, results from lack of ureteral bud formation [19].

Renal agenesis is a congenital condition in which an individual is born with the unilateral or bilateral absence of kidneys. Bilateral renal agenesis is incompatible with life, but those with solitary kidneys usually lead healthy lives and may not know of the condition unless found incidentally on imaging. Technological advances have created opportunities for prenatal diagnosis of renal agenesis through sonographic evaluation and color flow Doppler. Sonography allows the visualization of renal structures. If not located, the color flow Doppler is used to ascertain the blood flow of the renal artery [18]. In many cases of renal agenesis, the renal artery is absent [18]. Kidney donation and nephrectomies constitute the remaining portion of the population living with a solitary kidney. Nephrectomies are primarily performed as surgical management for renal carcinomas [20].

Upon dissection, it was evident that the left kidney and renal artery were absent, leaving only a solitary right kidney and right renal artery (Figure 4). Evidence of renal hypertrophy was noted by the following measurements of the kidney: 12.9 cm in length (superior to inferior pole), 8.2 cm in breadth (medial to lateral), and 5.6 cm from anterior to posterior. Standard kidney measurements average about 12 cm in length, 6 cm in breadth, and 3 cm in thickness [21]. Hypertrophy is a compensatory mechanism of the solitary kidney to increase the nephron filtration rate. Patients who undergo a nephrectomy have an acute increase in renal blood flow, leading to a cascade of events that ultimately initiates cellular activity, causing cell growth. With renal agenesis, hypertrophy occurs in utero via a poorly understood mechanism but is believed to be due to the renal nerve initiating nephrogenesis [22].

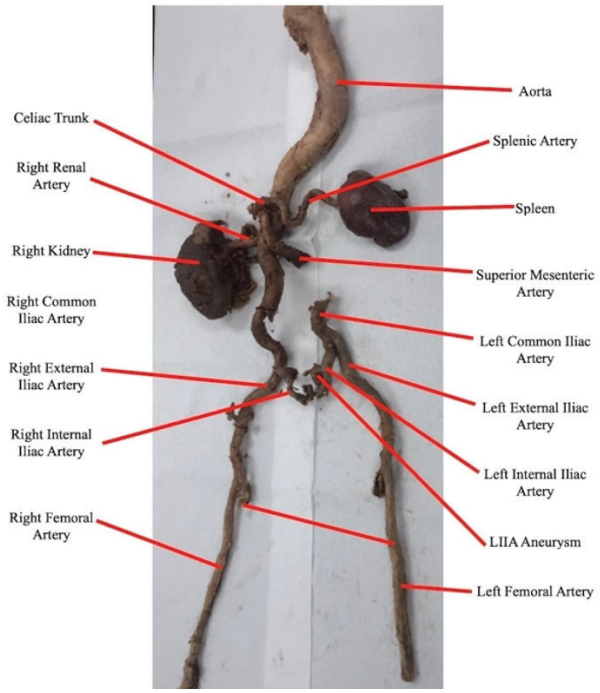

Figure 4) *Left and Right Common Iliac Arteries separated by Dissection (No evidence of a Left Renal Artery or Left Kidney).*

There was no indication of a left renal artery ever being present during dissection. During a nephrectomy, the renal artery is embolized, and a portion of it remains. The branch of the artery from the aorta is still present [23]. As this was not found in the cadaver, findings suggest that the variation could be the congenital form of solitary kidney—renal agenesis.

A literature review failed to find a case where both an IIAA and solitary kidney were present. Both conditions are rare, and finding both together is even more infrequent. Of note, a solitary kidney causes hypertension, which is a risk factor for IIAA formation—a potential source for the variations presented in this case [24].

Comorbidities

Although it has been stated one can live normally with only one kidney, there are comorbidities that, if untreated, can have negative impacts on the patient. Congenital and acquired solitary kidney conditions lead to similar comorbidities. Some of the leading complications are hypertension and end-stage renal disease [6].

Hypertension is a significant concern in this patient population. Congenital solitary kidney leads to decreased nephrons causing dysregulations in blood pressure. One study found that children with a congenital solitary kidney are at an increased risk for developing hypertension within their lifetime. It was suggested that even those who presented with blood pressure values within normal limits should be monitored periodically and receive long-term follow-up care [25]. Living donors who are otherwise healthy with no risk factors are not shown to be predisposed to hypertension after nephrectomy. However, elderly, obese, African American, and Hispanic donors are noted to be at increased risk for developing hypertension post-donation [26].

Donors were at higher risk of end-stage renal disease compared to those with two kidneys in similar demographics. However, the overall risk is considered low compared with the general population [6]. Compensatory hypertrophy allows about 85% of the glomerular filtration rate to

return to normal functioning in long-term follow-ups. Individuals with renal agenesis experience renal hypertrophy earlier in life. While those who have nephrectomies later in life exhibit a minimal change in glomerular pressure, patients with renal agenesis experience a 30% increase, leading to higher incidences of cardiovascular and renal disease. One study found that by age 18, children with a congenital solitary kidney exhibited signs of renal injury [22].

Management

Health care practitioners (HCPs) can assess for a congenital solitary kidney using ultrasonography. With more frequent prenatal ultrasound assessments, unilateral renal agenesis is being diagnosed more often. Although it is not a 100% effective diagnostic method, it allows HCPs to diagnose at increased frequencies and provide counsel and resources to parents [22]. Proteinuria, hypertension, and end-stage renal disease are possible complications that can arise later in life. If diagnosed, follow-up visits are recommended, and treatment of symptoms is indicated. Children with solitary kidney should not participate in contact sports to avoid injuring their kidney [18]. A review of various hospitals' post-operative instructions found that patients who undergo a radical nephrectomy can return to regular activity within three to four weeks [27-30].

Conclusion

We hypothesize that the presence of the IIIAA discovered in our male cadaver was likely attributable to the solitary kidney. It is well documented in the literature that individuals with a single functioning kidney are prone to developing hypertension, which is one of the major contributing risk factors of aneurysms. To our knowledge, this is the first time the pairing of these anatomical variations has been reported.

Acknowledgment

The authors are grateful to those who donate their bodies to science so that anatomical education and research might be performed. Results from research might increase overall knowledge and improve patient care. The donor and his family deserve our highest gratitude.

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