
ABSTRACT : Spontaneous peri-renal hemorrhage (SPH), also known as Wünderlich’s syndrome, is an uncommon and rare urologic emergency, which could become life-threatening, requiring immediate diagnosis and management. The diagnosis can be challenging, even with new imaging modalities, and management can vary from conservative approach, to surgical intervention. We present a case of jogging-induced SPH diagnosed using CT scan that was managed conservatively, with follow-up for up to five years using CT imaging studies.

Keywords : spontaneous peri-renal hemorrhage, Wünderlich, angiomyolipoma (AML), renal cell carcinoma (RCC), sub-capsular hematoma, idiopathic hemorrhage

INTRODUCTION

Spontaneous peri-renal hemorrhage (SPH) was first described in 1856 by Carl Wünderlich, a German physician, who identified a non-traumatic renal hemorrhage confined to the subcapsular and peri-renal space [1]. Most of the time, the clinical symptoms of SPH are non-specific (flank pain and/or abdominal mass), necessitating imaging studies. The overall sensitivity of CT scans in detection of SPH can reach up to 100% [2].

The most common etiologies are tumors, mainly angiomyolipoma (AML), followed by renal cell carcinoma (RCC). Among other etiologies are vascular entities such as polyarteritis nodosa and Wegener angeitis [1-3]. Few cases were labeled as idiopathic when the etiology could not be determined [4].

CASE REPORT

This is a case of a 42-year-old male, previously healthy, presented to Saint George Hospital University Medical Center (SGHUMC) in Beirut, Lebanon, in 2007, with a 3-day history of left flank pain, sudden in onset, after a single 30-minute jogging session, with no history of trauma, or repetitive violent sports activities. Pain increased in intensity over time and became unresponsive to conventional pain killers (paracetamol and non-steroidal anti-inflammatory drugs (NSAID)). On admission, the patient had left costo-vertebral angle (CVA) tenderness, with no evidence of overlying skin bruising. Laboratory workup revealed: hemoglobin (Hb): 13 g/dl; hematocrit (Hct): 38%; INR: 1.09. Urine analysis (U/A): red blood cells (RBC): 34; white blood cells (WBC): 4. Kidney, ureters and bladder X-ray (KUB) was normal.

An IV contrast uroscan study was done upon admission, revealing a hematoma, measuring 11 x 7 x 6 cm, with a density of 60-70 HU, arising from the posterior medullary region of the upper pole of the left kidney, not enhancing after IV contrast administration, causing the kidney to be slightly displaced anteriorly (Fig. 1).

The patient was managed with complete bed rest, serial complete blood count (CBC) and antibiotics (1 gm Cefizox IV q8hr). The patient had a gradual decrease in his Hb, reaching a level of 8.3, necessitating blood transfusion (three transfusions in total). A repeat CT scan was done on day 5, revealing and increase in the size of the hematoma, measuring 19 x 9 x 8 cm. An arteriogram done on day 5 after the CT came completely normal, with no vascular abnormality or active bleeding of the left kidney (Fig. 2). The patient was discharged after stabilization of his condition, Hb and Hct levels.

The CT imaging was repeated after three months (Fig. 3), and every six months thereafter for two years. The CT scan done two years after initial presentation
revealed gradual decrease in the hematoma, with no contrast enhancement, and no tumor was identified (Fig. 4). The patient continued to be followed with regular uroscan every year for up to five years, showing a decrease in the hematoma, and no evidence of renal tumor as an etiology explaining the SPH.

DISCUSSION

SPH was first mentioned by Wunderlich in 1856. Since then, and up till 1975, 78 cases of SPH were documented [3]. In addition, 165 extra cases were documented between 1985 and 1999 [2,4]. Both studies showed similar results regarding the etiology of the hemorrhage, as mentioned before.

Diagnosis can be challenging, especially with the non-specific clinical symptoms, making imaging studies a must when dealing with high suspicion of SPH [1]. In the acute phase of SPH due to tumor, the sensitivity of the CT scan is around 57% in identifying the cause of the hemorrhage. Ultrasonography (US) is not ideal in detecting the etiology of the hemorrhage, as it is challenging to differentiate solid mass from clotted blood [2]. Angiography is helpful in identifying vascular origins of the hemorrhage. MRI usage is controversial, but could be an alternative to CT when the latter is contraindicated.

It has been a common practice of most urologists to undergo surgical exploration facing a case of peri-renal hemorrhage, keeping in mind that renal tumors – mainly AML and RCC – are among the most common etiologies of SPH [3,5]. Complete inspection of the kidney in the presence of a hematoma can be quite challenging most of the time and could eventually results in nephrectomy, irrespective of the etiology. In his study, Zhang et al. [2]
identified 11 out of 165 cases of peri-renal hemorrhage to be of idiopathic origin, nine of which ended up with nephrectomy.

Compared to surgical exploration, conservative management can be of great value. Of notice, Belville et al. [6] stressed that in the face of a peri-renal hematoma, where the etiology of the hemorrhage cannot be determined, serial CT examinations and follow-up may present a viable alternative to surgical exploration, till a diagnosis is known, or resorption of hematoma. Baishya et al. [7] reported a case of SPH, which was treated conservatively, without surgical intervention. Post ureteroscopy SPH was reported by Bansal et al. [8] and was also managed conservatively. Sung et al. [9] reported a case of SPH caused by repetitive mild trauma injury due to hula-hooping, which was managed conservatively. It appears that conservative management of SPH – especially when the patient’s condition is stable, and imaging studies fail to demonstrate tumors such as AML or RCC as a cause of SPH – should be adopted, as opposed to immediate surgical exploration that most of the time would result in unnecessary nephrectomy [4, 6, 9].

Our case report, to the best of our knowledge, adds a couple of interesting information to the literature regarding SPH. First, it is the first of its kind to show a long follow-up period of a SPH – that was managed conservatively – for up to five years. All previous case studies had a relatively short follow-up periods, ranging from five days, to a maximum of 18 months [7, 9]. It is also the first of its kind to demonstrate SPH due to a single, non-violent activity of 30-minutes jogging, with no other etiology explaining the hematoma. The literature demonstrates several SPH cases due to sport activities, such as those reported by Sung et al. [9] and Fujita S. et al. [10]. However, the former was associated with heavy, daily, 30-minute-exercise using a hard plastic hoop, filled with iron beads, for an extended period of time, leading to repetitive mild injuries, and the latter was associated with martial arts kind of sports (judo), which has a high risk of severe blunt trauma injuries. In our case, there was neither any repetitive mild trauma, nor any kind of violent sport activity.

In conclusion, the usual approach facing SPH, which is surgical exploration that usually ends up with total nephrectomy, should be revised, especially in our kidney-sparing era. It is true that renal tumors continue to be the leading cause of SPH, which necessitates surgical intervention. However, when renal masses cannot be identified, and the patient is hemodynamically stable, conservative management should be given priority over surgical intervention. But in order to embrace such an approach, more accurate techniques for diagnosis of the etiologies of the hemorrhage are needed. Of them all, CT scan, triphasic, with thin slices (≤ 5mm) seems to offer the best diagnostic method. As such, serial usage of CT scans in the process of follow-up is highly justified and even recommended to avoid unpleasant sequelae, especially with the ongoing trend towards kidney-sparing strategies.

REFERENCES