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Original Research

A preliminary study on classification and therapeutic strategies for spontaneous perirenal hemorrhage

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ABSTRACT

Background: The aim of our study was to report our experience in the classification and therapeutic management strategies for spontaneous perirenal hemorrhage (SPH).

Methods: From September 2005 to April 2015, 20 patients with SPH were newly diagnosed in our hospital. Their clinical features, image findings, identification of underlying causes, and therapeutic management were retrospectively analyzed, and relevant literature was reviewed. In this study, patients were classified according to the degree of severity of the disease or emergency imaging diagnosis of underlying causes. On the basis of the former, patients were classified as critical and noncritical, and on the basis of the latter, patients were classified as renal cell carcinoma (RCC), undefined solid neoplasm, angioleiomyolipoma (AML), and unknown cause.

Results: In the acute stage, contrast-enhanced computed tomography (CT) was superior to ultrasonography for both diagnostic accuracy of SPH ($p = 0.02$) and etiology discovery power ($p = 0.004$). The results of contrast-enhanced magnetic resonance imaging (MRI) were identical to those of contrast-enhanced CT. We summarized a flowchart in the whole classification and therapeutic strategies of SPH. According to the imaging diagnosis of underlying causes, all the patients with undefined solid neoplasm or RCC underwent emergency operation. Patients with AML or unknown cause underwent selective arterial embolization (SAE) or conservative management according to the critical degree. Acute hemorrhage was controlled in 19 cases, of which 14 were cured by the operation and only one critical patient with severe shock died shortly despite rescue efforts.

Conclusions: Contrast-enhanced CT or MRI is the first choice of imaging examination, which could not only accurately diagnose SPH but also detect the underlying causes. Choice of therapeutic strategies for SPH should vary according to the identification of critical patients and imaging diagnosis of underlying cause.

1. Background

Spontaneous perirenal hemorrhage (SPH) is a relatively uncommon but often diagnostically challenging condition. Carl Wunderlich is credited with the first clinical description of Wunderlich syndrome (WS) in 1856, as SPH was confined to the subscapular and perinephric space in patients with no history of trauma. The incidence of this disease is low, and most of the literature are case reports [1,2]. Usually, patients with SPH have potential pathological changes of the kidney. Renal neoplasms are the most common causes of SPH, which account for 60%–65% of all cases. Renal angioleiomyolipoma (AML) is the most common benign neoplasm responsible for SPH (accounting for 30%–40%), whereas renal cell carcinoma (RCC) is the most common malignant neoplasm (accounting for 25%–35%). SPH caused by other neoplasms such as nephroblastoma, nephrosarcoma, urothelial carcinoma, and renal oncocytoma is rare [1–3]. Vascular diseases are the second common cause of SPH, which account for 20%–30% of SPH cases, in which polyarteritis nodosa accounts for half of the vascular pathology. Other vascular etiologies include renal aneurysm, arteriovenous malformations (AVMs), arteriovenous fistulas (AVFs), and so on [4,5]. Furthermore, other underlying causes of SPH such as hereditary and acquired renal cystic diseases as well as infectious disease (suppurative renal disease and pyelonephritis) were also reported [2,3].

SPH is typically characterized by acute flank pain, hypovolemic shock, hematuria, and flank mass. The onset of this disease is abrupt, and it causes excessive bleeding, thereby leading to a quick hypovolemic shock. Owing to massive retroperitoneal bleeding in patients, the underlying causes of SPH could not be revealed in around 20% of the cases by emergency imaging findings, and this makes it difficult to choose an optimal management. As the majority of the underlying

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etiologies causing SPH are benign diseases such as AML, vascular diseases, and renal cyst, it is therefore important to avoid unnecessary nephrectomy, and standard procedures might contribute to diagnosis and management of SPH.

In this study, we retrospectively analyzed and evaluated the outcomes of various diagnostic imaging modalities and different management approaches in 20 patients with SPH treated in our hospital, and relevant literature was reviewed. We proposed a simple and effective flowchart for SPH therapeutic management, and this might facilitate physicians to select an optimal management and ensure maximum benefit for patients.

2. Patients and methods

2.1. Patient information

We retrospectively collected information of 20 patients who were confirmed as having SPH between 2008 and 2015 at a hospital. The Clinical Research Ethics Committee at our institute had approved the study and granted permissions to access the patient records. This work has been reported in line with the STROCSS criteria [4]. Patients' demographics are described in Table 1. The median age of the patients was 53 years (range, 14–83 years). Among the 20 patients, 11 cases were initially confirmed in our hospital and nine cases were transferred from other hospital. None of these patients had a history of trauma or previously any anticoagulant therapy.

All patients presented with acute flank or abdominal pain. Six cases displayed hypovolemic shock. Among them, five cases had moderate shock (systolic blood pressure ≤ 90 mmHg) and one patient had severe shock (systolic blood pressure ≤ 60 mmHg). All the patients had percussion pain in the renal region and only one patient had ecchymosis on the flank and back. There were no signs of peritoneal irritation such as obvious tenderness, rebound tenderness, and muscle rigidity in all cases.

2.2. Laboratory examination

Blood routine tests, urine routine tests, liver function, renal function, and coagulation function were performed in all cases. Ten patients presented with anemia, including six cases with severe anemia (Hb < 60 g/L) and four cases with mild-to-moderate anemia. Five cases had coagulation abnormalities and two cases had microscopic hematuria.

2.3. Imaging examination

Color Doppler ultrasound and contrast-enhanced computed tomography (CT) were performed in all patients. Contrast-enhanced magnetic resonance imaging (MRI) was performed in five patients. On color Doppler ultrasound, SPH with acute hematoma appeared as an isoechoic or hyperechoic subcapsular or perinephric collection, thus causing compression and displacement of renal parenchyma. Blood-rich mixed masses with hyperechoic fat component ingredient indicated an AML. On contrast-enhanced CT, SPH with acute hematoma was categorized as those occurring in renal subcapsular space, perirenal space, and pararenal space according to the scope of hematoma; its CT values varied from 60 to 80 HU (Hounsfield units). As hemorrhagic lesions contain mixture of density masses with fat density (CT value ≤ −20 HU), imaging diagnosis indicated rupture of AML (Fig. 1 A, B). During the corticomedullary phase of contrast-enhanced CT, heterogeneous enhancing hypervascular soft tissue mass within the hematoma indicates rupture of RCC (Fig. 1 C, D). On contrast-enhanced MRI, relative to the muscle, acute hematoma (< 7 days) showed equal or slightly low signal intensity on I, which stands for “image,” and low signal intensity on II, which stands for “image. If the predominant component of hematoma was fat, it could present with a high signal on I, which stands for “image,” and slightly high on II, which stands for “image,” and displayed a low signal on the fat-suppressed sequence of I, which stands for “image” and II, which stands for “image.” This evidence indicated the rupture of AML.

2.4. Treatment and follow-up

In our study, considering the severity of hemorrhagic shock caused by SPH, the critical degree of a patient was judged on the basis of blood pressure and shock index, which indicates moderate shock. Meanwhile, kidneys, located in the retroperitoneum, cause a relatively lower spped of blood loss than abdominal organs; therefore, the hemoglobin level can be a reliable indicator of the severity of blood loss. Critical patients with SPH are defined as patients with systolic blood pressure ≤90 mmHg, shock index ≥1.5, and Hb level < 60 g/L. Six critical cases were then diagnosed. The shock index was separately 2.2, 1.7, 1.6, 1.6, 1.8, and 1.5. The hemoglobin level was separately 29 g/L, 46 g/L, 51 g/L, 52 g/L, 42 g/L, and 58 g/L. Management of patients with hemorrhagic shock was by deep venous catheterization, monitoring of vital signs, shock index and urine volume, fluid resuscitation,

Table 1
Characteristics of patients with WS.

No.	Gender	Age	Critical patient	Imaging diagnosis of underlying cause	Management	Confirmed etiology
1	F	43	YES	AML	Transferred from other hospital, emergency RN	AML
2	M	64	YES	AML	Transferred from other hospital, died after transient rescue efforts	AML
3	F	60	NO	AML	Conservative management, selective PN	AML
4	F	43	NO	AML	Conservative management, selective RAPN	AML
5	F	53	NO	AML	Conservative management, selective RAPN	AML
6	M	35	NO	AML	Conservative management	AML
7	M	73	NO	AML	Conservative management	AML
8	F	66	YES	RCC	Emergency RN	RCC
9	M	14	NO	RCC	Emergency RN	RCC
10	F	52	YES	Renal metastasis from lung cancer	Emergency SAE	Renal metastasis from lung cancer
11	F	63	NO	Undefined solid neoplasm	Emergency RN	RCC
12	F	40	NO	Undefined solid neoplasm	Emergency PN	Complex renal cyst
13	M	54	YES	Unknown cause	Emergency SAE	Renal cyst
14	M	23	NO	Unknown cause	Conservative management, selective PN	AML
15	M	83	NO	Unknown cause	Conservative management	Renal cyst
16	F	38	YES	AML	SAE invalid, emergency RN	AML
17	F	53	NO	AML	Conservative management, selective PN	AML
18	F	28	NO	AML	Conservative management, selective PN	AML
19	F	59	NO	AML	Conservative management, selective RAPN	AML
20	F	60	NO	AML	Conservative management, selective RAPN	AML