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Research Article

Spontaneous Perinephric Hematoma in Patients of Pyelonephritis: Case Series of an Uncommon Complication

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Abstract

Introduction: Spontaneous Perinephric Hematoma (SPH) used to remain undiagnosed because of deep-seated location of kidney protected by multiple anatomical envelopes. This is no more the case after easy availability of ultrasound and CT scan; and now, SPH is being diagnosed more often. SPH can occur due to various causes like inflammatory, infective, vascular conditions and bleeding diathesis. SPH can occur in association with pyelonephritis, whether non-obstructive or due to ureteric obstruction. There is no established protocol for management of SPH associated with pyelonephritis. In an attempt to fill up this vacuum, we evaluated our cases of SPH which occurred in patients admitted for indoor treatment of non-obstructive pyelonephritis with the aim was to arrive at appropriate management protocol of SPH associated with pyelonephritis. *Methods*: A review of record files of 82 cases of non-obstructive pyelonephritis admitted in department of urology of Goa Medical College, GOA, India from January, 2020 to April, 2024 was done. SPH was detected in 5 cases, and as per our protocol, in all 5 cases, SPH was treated by minimally invasive approach in the form of Single Stage Aspiration (SSA) +/- Pigtail catheter placement. Out of 5 cases, 2 patients were cured with SSA alone; 1 patient required an additional procedure in the form of 2nd stage Pigtail placement due to recurrence of hematoma after SSA; and 2 cases were treated with SSA + Pigtail done in same sitting. No emergency exploration of SPH was done in any case and renal salvage could be achieved in all 5 cases. *Conclusion*: SSA +/-Pigtail Catheter as primary therapeutic modality was found to be safe and effective for treatment of SPH in Pyelonephritis though the number of cases in this study was small.

Keywords

Perinephric Hematoma, Wunderlich Syndrome, Pyelonephritis, Lenk's Triad, Spontaneous Perinephric Hematoma

1. Introduction

Etiology of perinephric hematoma can be categorized as i) Traumatic (Biopsy, ESWL & Trauma), ii) Neoplastic with AML in 50%, and iii) Inflammatory, Infective & Vascular conditions [1]. In the absence of h/o trauma or an iatrogenic cause, the condition is called SPH [2], also known as 'Wunderlich Syndrome' named after a German physician, Carl August Wunderlich who, in 1856, described the condition as "Spontaneous Renal Capsule Apoplexy" as hemorrhage in subcapsular or perirenal space without prior h/o trauma [3, 4]. Before routine availability of ultrasound and CT scan, SPH was considered a rare entity, but with the availability of current imaging modalities, SPH is rare no

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more.

In a case of pyelonephritis, SPH is picked up initially on routine ultrasound which shows hyperechogenicity in the vicinity of kidney. CT, being a remarkable modality to delineate the anatomy of kidney by virtue of multiple fat-containing envelopes around the kidney, detects SPH more often than what we suspected earlier. In the changing scenario, we need to define the management protocol for SPH. *"Hands-off"* approach risks the kidney function; and exploration of SPH in the setting of pyelonephritis almost always ends up in nephrectomy and carries mortality to the tune of 50% [5]. We present a series of 5 cases of SPH which were detected amongst 82 patients of non-obstructive pyelonephritis admitted in our department and treated safely.

2. Material & Methods

It was a retrospective analysis and record files of all patients admitted with diagnosis of Pyelonephritis in Dept. of Urology at Goa Medical College, GOA, INDIA from Jan, 2020 to Apr, 2024, were reviewed.

There were 82 patients of Non-obstructive Pyelonephritis and out of these, 22 had perinephric collection which was hemorrhagic in 5 patients as reported by radiologist after ultrasound and CT scan. 17 cases in which there was clinical/radiological evidence of infected collection did not form part of the study. Remaining 5 patients i.e. perinephric hematoma had undergone therapeutic aspiration +/-Pigtail under ultrasound guidance.

3. Results

In 2 out of 5 patients, aspirate was sanguino-purulent although there was no previous clinical/radiological sign of infection; hence a pig-tail was placed in these 2 cases in the same sitting. In remaining 3 patients, near total aspiration and decompression of hematoma could be achieved with SSA. In one out of these 3 cases, recurrence of hematoma was found on follow-up ultrasound done on 10^{th} day and this patient required a 2^{nd} stage pig-tail placement although satisfactory decompression had been achieved at SSA.

Further analysis of clinical data of these 5 cases revealed that all of them were above 50 years in age, 3 were female and 2 males. 3 Patients, all female, were having Type II DM. All 5 patients had presented with flank pain which was associated with fever in 4 cases. There was h/o hematuria in 3 patients; asthenia was present in 4 cases; palpable lump in one case and urine was turbid in one case. There was one patient who was on anti-platelet therapy, but his coagulation profile reported no abnormality.

4. Discussion

Classical presentation of SPH is described as Lenk's Triad i.e. acute flank pain, flank mass & hypovolemic shock, but it is seen in a small percentage of patients and the condition presents with a multitude of symptoms ranging from pain abdomen to hypovolemic shock [6]. Symptoms due to SPH associated with upper urinary tract infection (UTI) are likely to be masked by those due to pyelonephritis and since there is no aggressive bleeding in this situation, these cases are not expected to produce hypovolemic shock which is typical of bleeding due to tumor or trauma, our series deviated from classical presentation. Only one patient presented with flank mass and none of 5 patients was in shock possibly because all these 5 patients were in-door and perinephric hemorrhage was picked up early by ultrasound and attended.

SPH is initially picked up on ultrasound as an avascular iso-echoic or hyperechoic area in the vicinity of kidney in early stage and may be enveloping the reniform kidney, and hypoechoic to anechoic heterogenous fluid collection with internal septation which may cause compression of subjacent renal parenchyma [6]. Ultrasound findings are then confirmed on CT, the latter having higher diagnostic accuracy ($p=0.02$) and etiology diagnostic power ($p=0.004$) [7]. Ultrasound should not be used to gauge the etiology of SPH whereas accuracy of CT to pick up etiological factor is 52% [2]. Sensitivity of CT for detecting blood in perinephric space is 100% [2]. Belville *et al.* compared ultrasound, CT and angiography for the diagnosis of perinephric hematoma and found that CT is the most valuable diagnostic modality for suspected perinephric hematoma [8]. Dual energy CT can diagnose PN hematoma even without contrast, but contrast enhancement is still desirable to pick up possible enhancing lesion as a cause of hematoma and also to look for continuing bleeding into hematoma [6]. Contrast-enhanced MRI is only as good as CECT for diagnosing SPH [2, 7]. Renal angiography+/-embolization is indicated in SPH if (1) there is solid renal mass to identify angiomyolipoma, (2) there is evidence of active bleeding as in cases of coagulopathy and (3) patient is hemodynamically unstable [7]. Perinephric hematoma occurring around infected kidney is different from perinephric hemorrhage caused by trauma, procedure or neoplasm because there is no sudden large extrusion of blood into perinephric space as it would have occurred in other conditions involving damage to a sizeable vessel, and it does not extend into retroperitoneum unless infected.

In their largest study involving 165 patients over 47 previous studies, Zhang *et al.* had found neoplastic etiology to be the commonest causative factor for SPH, and infection constituted 1.2% of all cases of perinephric hematoma including those caused by trauma [1]. But, this meta-analysis included large number of studies done before easy availability CECT for evaluation of upper UTI. In a more recent study, pyelonephritis was found to be the causative factor in 4 out of a total of 28 cases of SPH [9]. Shah *et al.* reported the renal infections to be contributing to 5-10% cases of SPH [6]. In our study, we have considered SPH occurring in the setting of non-obstructive pyelonephritis only. This review

analysis shows that SPH occurs in elderly having pyelonephritis and there is preponderance for SPH in diabetics [10]. Pyelonephritis in 3 diabetic female patients in this study suggests that upper UTI was due to retrograde entry. Renal infection in diabetes is a predisposing factor for SPH because infection-associated parenchymal necrosis and coagulation of microvasculature results in bleeding in subcapsular/perinephric space [6]. Hence, poor response to antibiotic therapy in a diabetic patient having upper UTI should raise suspicion and one should start looking for associated entities like SPH.

A number of case reports are there in the literature where SPH occurred following initiation of anticoagulation. But, coagulopathy does not seem to be a pre-requisite for development of SPH because only 1 patient out of 5 was on aspirin and there was no evidence of coagulopathy in any patient. Possibly, it is aggressive anticoagulation which is likely to enter into causative list of SPH as it occurred after combined rivaroxaban and clopidogrel in the case reported by Lee *et al.* where they managed the case by angioembolization [11]. In their case, angiography was a logical investigation because CECT had shown extravasation of contrast into perinephric space. In the setting of possible infection, perinephric hematoma is a common event with ureteric obstruction due to stone or following ureterorenoscopic intervention [12]. Our case series does not represent exact picture of SPH in pyelonephritis because some of the patients of SPH due to pyelonephritis would have come to emergency department in septic shock and got admitted in critical care under the charge of physician and managed in medicine unit due to accompanying co-morbidities and did not involve active urosurgical intervention. Such cases would have escaped the retrospective review of urology files. Secondly, our review did not include cases of SPH where concomitant ureteric obstruction was there.

Rather than resorting to hazardous surgical intervention in the form of exploration for SPH associated with pyelonephritis, this study reveals that minimally invasive procedure like ultrasound guided SSA+/-pigtail placement is safe and effective and we advocate this approach which should be exercised proactively. 2 out of 5 patients in this series recovered with SSA alone; and 1 case responded to SSA+2nd stage pigtail. 2 patients required pigtail at initial attempt with SSA. But, it is worth mentioning that all 5 patients survived the emergency; and emergency exploration with possibility of high risk nephrectomy was avoided. Renal salvage in all 5 cases was an additional gain.

The combined expert panel of World Society of Emergency Surgery and American Association for the Surgery of Trauma (WSES-AAST) recommended early intervention primarily in the form of angioembolization in case of peri-renal hematoma [13]. But, here we are managing a non-traumatic condition. Angioembolization is likely to be beneficial when we expect an actively bleeding vessel as in case of trauma [13] or vascular condition like polyarteritis nodosa [5]. Hematomas in our cases were found in the setting of pyelonephritis and there was no suspicion on CT or documented evidence of a bleeding vessel. Hence, we did not do renal angiography. Chung *et al.* in their isolated case report have mentioned DJ stent-induced perinephric hematoma and its management by tube drainage by interventional radiologist without untoward event [12]. They also recommended limiting the procedure to aspiration in 1st sitting if tube drainage was avoidable although their isolated case did require 2nd stage procedure and tube drainage. The literature recommends indications of early percutaneous intervention in perinephric hematoma in case of (1) interactable pain, (2) possible infection of hematoma and (3) renal compression and ischemia of kidney [14].

Contrary to our series, when SPH is encountered in association of solid renal neoplasm, there is no role of conservative treatment and emergency nephrectomy is required [7].

Some authors have described spontaneous resolution of isolated SPH over a period of 2 years with restoration of anatomy of kidney, but they did not do physiological studies to find out residual renal function after SPH had resolved in their case [15]. We don't buy this approach especially in pyelonephritis where encysted hematoma surrounding an infected kidney & harboring microbes, always runs the risk of getting converted into an abscess as evident from sanguino-purulent aspirate in 2 out of 5 cases in this series. Nawaj *et al.* also recommended conservative approach in stable patient [16]. But, their patient was a case of perinephric bleed caused by anticoagulation rather than an infective process. Kamarudin *et al.* managed their case of perinephric hematoma conservatively, but their case can't be classified as SPH because hematoma had followed trauma caused by biopsy needle [17]. Nakashima *et al.* described successful conservative management of SPH due to Microscopic Polyangiitis (MPA) contrary to aggressive management of SPH due to Polyarteritis nodosa (PAN) on the ground that bleeding due to MPA is very slow as compared to aggressive bleed in PAN [18]. But, *'Hands-off'* approach in SPH does not find support in the literature because it can produce *'Page'* kidney and life-long hypertension [6].

Nomikos reported management of perinephric hematoma caused by DJ stent in a case of ureteric obstruction and they successfully treated the patient by supportive means [14]. They also remained watchful for evidence of any aggressive collection or infection for which they were prepared to do intervention which was never required. We find no wrong in this approach as long as the renal unit remains uninfected. Petros *et al.* reported a case of SPH caused by forniceal rupture where they were compelled to go for surgical intervention because interventional radiologist expressed inability to tackle the situation by tube drainage due to logistical reasons [19]. As usual, aggressive intervention ended in nephrectomy where a possibility of renal salvage was there. In the literature, robot-assisted intervention for SPH has started appearing, but this approach needs to be subjected to the test of

time [20]. Zuckerman *et al.* reported a case of SPH due to upper polar infarct [10]; neither the conservative approach nor our minimally invasive approach is going to work in this kind of serious situation and their decision to intervene followed by nephrectomy was the most appropriate one.

5. Conclusions

SPH associated with pyelonephritis can be safely and effectively managed by minimally invasive procedures like SSA+/-Pigtail Catheter placement and emergency exploration of hematoma is unwarranted.

Abbreviations

SPH	Spontaneous Perinephric Hematoma
SSA	Single Stage Aspiration
AML	Angiomyolipoma
UTI	Urinary Tract Infection
WSES	World Society of Emergency Surgery
AAST	American Association for Surgery of Trauma
MPA	Microscopic Polyangiitis

Statement of Ethic

This study was a retrospective observational review of records of patients treated in our hospital on earlier dates and no prospective intervention was involved. The procedures involved were standard scientific minimally invasive ones and all these procedures were applied to established absolute indications of intervention. Procedures were carried out after following standard procedure of "Written Informed Consent" by the patient as routinely applicable to invasive procedures in our institution. For academic purpose, permission of Institutional Review Board was obtained and waiver of consent for publication from individual patient was granted as patient's identity was not disclosed for data analysis or publication.

Author Contributions

Ekansh Gupta: Review of Literature and Manuscript writing

Madhumohan Prabhudessai: Supervision as Head of Urology

Rajesh Halarnakar: Did the procedure in 2 cases

Cardoso Amanda: Operated on 2 patients

Prashant Lawande: Did 2 procedures in 1 case

Kartik Shetty: Data collection, Review and Editing

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Data Availability Statement

The above data has been compiled from the records of patients admitted in the Department of Urology at public-funded Goa Medical College, GOA, INDIA and it remains the property of Government of Goa. The record files belong to Department of Urology and are maintained with Medical Records Department of the Goa Medical College.

Conflicts of Interest

The authors declare no conflicts of interest.

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