Osmangazi Journal of Medicine e-ISSN: 2587-1579

Ten-Year Experience with Atraumatic Splenic Rupture in a Tertiary University Hospital

Üçüncü Basamak Bir Üniversite Hastanesinde Atravmatik Dalak Rüptürü ile İlgili On Yıllık Deneyim

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Ethics Committee Approval: This study was approved by the Ethics Committee of the Eskisehir Osmangazi University (Decision no: 15, Date: 03.10.2024).

Informed Consent: The authors declared that informed consent form was signed by the patient.

Authorship Contributions: All authors contributed equally to the study.

Copyright Transfer Form: Copyright Transfer Formwas signed by all authors

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

> Received : 27.11.2024 Accepted :06.01.2025 Published: 07.01.2025

Abstract: In this present study, we aim to share our clinical experience in patients with atraumatic splenic rupture (ASR). The records of all patients were reviewed from January 2015 to May 2024, retrospectively. We had 8 patients diagnosed with ASR. Of the patients 62.5% (n=5) were female. The median age was 52.5 (37-72) years. Three patients had a history of malignancy, two had hematologic disease, and three had autoimmune disease. Two patients had previously undergone abdominal surgery. Two patients had a history of oral anticoagulant use. The median onset of symptoms was 16 (1-168) hours. Splenomegaly was detected in three patients via computed tomography. In one patient, a distal pancreatectomy was performed in addition to splenectomy, while for another patient, distal pancreatectomy, gastric wedge resection, and packing were performed. The median operation time was 122.5 (60-210) min. The median hospital stay was 8.5 (1-41) days. Pathological results showed adenocarcinoma metastasis in one patient (due to endometrial cancer), and necrotising granulomatous inflammation in one patient (due to abdominal tuberculosis). Early postoperative complication was seen only one patient as pancreatic fistula. Mortality occurred in three patients. One patient died in the second postoperative hour from hypovolemic shock, another from sepsis, and the third from a subarachnoid hemorrhage. Despite its clinically vague presentation, the diagnosis of spontaneous splenic rupture should be considered in patients with no history of trauma but who present with hypovolemic shock, acute abdomen or abdominal pain of unknown etiology.

Keywords: Acute Abdomen, Spontaneus Spleen Ruptur, Nontraumatic Splenic Damage, İntraabdominal Hemorrhage

Özet: Bu çalışmada, atravmatik dalak rüptürü (ASR) olan hastalardaki klinik deneyimimizi paylaşmayı amaçlıyoruz. Tüm hastaların kayıtları Ocak 2015'ten Mayıs 2024'e kadar retrospektif olarak incelendi. ASR tanısı konulan 8 hastamız vardı.. Hastaların %62.5'i (n=5) kadındı. Ortanca yaş 52.5 (37-72) yıldı. Üç hastada malignite öyküsü, iki hastada hematolojik hastalık ve üç hastada otoimmün hastalık vardı. İki hasta daha önce karın ameliyatı geçirmişti. İki hastada oral antikoagülan kullanma öyküsü vardı. Semptomların başlangıcı ortanca 16 (1-168) saatti. Üç hastaya Epstein-Barr Virüsü ve Sitomegalovirüs testleri yapıldı ve tümünde sonuç negatifti. Üç hastada bilgisayarlı tomografi ile splenomegali tespit edildi. Bir hastada splenektomiye ek olarak distal pankreatektomi yapılırken, bir diğer hastaya distal pankreatektomi, gastrik wedge rezeksiyon ve packing yapıldı. Operasyon süresi ortanca 122.5 (60-210) dakikaydı. Ortanca hastanede kalış süresi 8.5 (1-41) gündü. Patolojik sonuçlar bir hastada adenokarsinom metastazı (endometriyal kanser nedeniyle) ve bir hastada nekrotizan granülomatöz inflamasyondu (abdominal tüberküloz nedeniyle). Ameliyat sonrası erken komplikasyon sadece bir hastada pankreas fistülü olarak görüldü. Üç hastada mortalite görüldü. Bir hasta postoperatif ikinci saatte hipovolemik şoktan, bir diğeri sepsis nedeniyle ve üçüncüsü subaraknoid kanama nedeniyle öldü. Klinik olarak belirsiz sunumuna rağmen, travma öyküsü olmayan ancak hipovolemik şok, akut karın veya etiyolojisi bilinmeyen karın ağrısı ile gelen hastalarda spontan dalak rüptürü tanısı düşünülmelidir.

Anahtar Kelimeler: Akut Karın, Spontan Dalak Rüptürü, Travmatik Olmayan Dalak Hasarı, İntraabdominal Kanama

How to cite/ Atıf için: Zengin A, Yurdakul MM, Angın YS, Ulaş M, Ten-Year Experience with Atraumatic Splenic Rupture in a Tertiary University Hospital, Osmangazi Journal of Medicine, 2025;47(1):128-132

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1. Introduction

Atraumatic splenic rupture (ASR) is rare but lifethreatening, with a high risk of mortality, making it a critical emergency (1). In cases of acute abdominal pain without a history of trauma, ASR is often overlooked in the differential diagnosis (2).

In the literature, rupture without a history of trauma is commonly referred to as "spontaneous" "atraumatic" rupture (3). However, this terminology can be misleading. In 1991, Crate and Payne (4) defined spontaneous splenic rupture as occurring in the absence of trauma or exertion, without any underlying disease affecting the spleen, and with no evidence of prior splenic trauma or rupture (such as adhesions or scarring). Furthermore, their criteria required that the spleen appear normal upon macroscopic and histological examination and that be no findings suggesting acute convalescent phase antibody titers linked to viral infections affecting the spleen. Thus, while all spontaneous splenic ruptures fall within the ASR category, not all ASR cases qualify as spontaneous ruptures. Therefore, in this paper, we will refer to cases that other authors may have labeled as "spontaneous splenic rupture" under the umbrella term ASR.

This study aims to identify the characteristics, etiology, and diagnostic and therapeutic processes in our patients with ASR, as well as to highlight our approach to these less commonly encountered cases.

2. Materials and Methods

This study was approved by the local ethics committee (2024/15). Inclusion criteria were age >18 years and emergency splenectomy due to splenic rupture without a history of trauma. Patients who underwent elective open/laparoscopic splenectomy and splenectomy due to iatrogenic splenic injury during other surgeries were excluded from the study. Ultimately, eight patients met the criteria and were included in the study. A length of 12 to 20 cm is indicative of splenomegaly (measurements by ultrasonography (USG) or computed tomography (CT) (5).

Age, gender, The American Society of Anesthesiologists classification (ASA), comorbities, previous surgery history, oral anticoagulant use history, onset of sign, hemoglobin (Hb), hematocrit (Htc), white blood cell (WBC), lymphocyte, platelet (Plt), international normalized ratio (INR), amylase, viral marker (Epstein-Barr Virus (EBV),

Cytomegalovirus (CMV) levels, imaging methods, presence of splenomegaly as radiologically, operation type, combined resection, hospital mortality, and pathological results were analyzed.

Statistic

Continuous variables are reported as medians (range), and categorical data are presented as frequencies and percentages.

3. Results

A total of 285 patients underwent splenectomy, with or without additional organ resection, between January 2015 and May 2024, and 8 patients were diagnosed with ASR.

Table 1 shows the demographic data and preoperative biochemical test results of the patients. Of the patients, 62.5% (n=5) were female. The median age was 52.5 (37-72) years. One patient had secondary metastatic neoplasia (endometrial cancer), one had a malignant hematologic disease (acute myeloid leukemia [AML]), and one had a primary neoplastic disease (mesothelioma). Two patients had non-malignant hematologic conditions (disseminated intravascular coagulation [DIC] and idiopathic thrombocytopenic purpura [ITP]), while three had a history of autoimmune disease (systemic lupus erythematosus [SLE], ankylosing spondylitis, and Crohn's disease). No underlying cause was identified in two patients.

Two patients had history of prior abdominal surgery (appendectomy and laparoscopic cholecystectomy), and two patients had history of oral anticoagulant use. The median onset of symptoms was 16 (1-168) hours. Patient 5 presented to the hospital with a three-week history of dyspnea. A CT scan revealed a splenic laceration incidentally. Patient 6's delayed diagnosis was due to the patient's hospitalization in the hematology ward at the time of consultation and the very late onset of symptom. EBV and CMV tests were conducted on three patients, and results were negative in all cases.

Diagnosis methods and follow-up outcomes of the patients are shown in Table 2. Splenomegaly was detected for three patients in CT. In Patient 5, a distal pancreatectomy was performed in addition to a splenectomy, while for Patient 7, distal pancreatectomy, gastric wedge resection, and packing were performed. The others were performed only splenectomy. The median operation time was

122.5 (60-210) minutes. The median hospital stay was 8.5 (1-41) days. Pathological results showed adenocarcinoma metastasis in one patient (due to endometrial cancer), and necrotising granulomatous inflammation in one patient (due to abdominal tuberculosis), the others pathological results were reported normal spleen tissue.

Patient 8 underwent two reoperations. The first was for atraumatic splenic rupture, during which splenectomy, distal pancreatectomy, gastric wedge resection, and packing were performed. On the second postoperative day, the patient was reoperated for depacking. An early postoperative complication included a pancreatic fistula, for which interventional radiology placed an external stent to manage the leakage. However, on the 22nd postoperative day, abdominal bleeding occurred, necessitating a third surgery. No focus detected.

Mortality occurred in three patients on postoperative 0th (Patient 4), 8th (Patient 3), and 20th (Patient 6) day. Patient 4 died in the second postoperative hour from hypovolemic shock, Patient 3 died of sepsis, and Patient 2 died from a subarachnoid hemorrhage.

Table 1. Demographic data and preoperative biochemical test results of the patients

Patient number	P-1	P-2	P-3	P-4	P-5	P-6	P-7	P-8
Age (year)	54	50	38	60	68	72	51	37
Gender (F/M)	F	M	F		F	F	M	M
ASA	2	2	2	2	3	2	2	2
Comorbidities	Endometrial	Ankylosing	Mesothelio	SLE, ITP	HT, CAD	AML	CRD	Chron
	cancer, DIC,	spondylitis	ma					diseas
	Asthma							e
Previous surgery	Appendectomy	-	Left	Thyroidec	Laparosco	-	-	-
			pneumonuct	tomy	pic			
			omy		cholecyste			
					ctomy,			
					menengio			
					ma			
-					resection			
Oral anticoagulant	+	-	-	-	+	-	-	-
use history (+/-)								
Onset of sign (hour)	6	72	16	1	incidental	168	8	28
Hb (g/dl)	8.5	6.5	5.9	4.5	8.5	5	6.7	8.8
Htc (%)	26.6	28.8	17.7	13	24.6	14.7	19.4	25.7
WBC $(10^3/\text{uL})$	27800	13620	22200	39900	23030	530	26070	24720
Lymphocyte (/mm ³)	2300	2530	400	3900	1130	280	2120	1060
Plt ((/mm ³)	391000	204000	90000	21000	389000	20000	71000	29700
								0
INR	1,34	1,22	1,30	1,26	1,31	1,17	1,4	1,07
Amylase (U/L)	34	59	12	56	43	25	142	49
Viral markers	NA	negative	NA	NA	NA	NA	negati	negati
(CMV/EBV)							ve	ve

F: Female, M: Male, The American Society of Anesthesiologists classification, DIC: Disseminated intravascular coagulation, HT: Hypertension, SLE: Systemic lupus erythematosus, ITP: Idiopathic thrombocytopenic purpura, CAD: Coronary artery disease, AML: acute myeloid leukemia, CRD: chronic renal disease, Hb: Hemoglobin, Htc: Hematocrit, WBC: White blood cell, Plt: Platelet, INR: International normalized ratio EBV: Epstein-Barr Virus, CMV: Cytomegalovirus, NA:nonavailable

Patient number	P-1	P-2	P-3	P-4	P-5	P-6	P-7	P-8
Imaging methods	CT	USG, CT	USG, CT	CT	CT	USG, CT	USG, CT	CT
Splenomegaly (+/-)	-	-	-	-	+	+	+	-
Operation type	Splenectom y	Splenectom y	Splenectomy	Splenectomy	Splenectomy + Distal pancreatecto my	Splenectom y	Splenectomy+ Distal pancreatectomy + gastric wedge resection+ Packing	Splenecto my
Hospital mortality	-	-	Po 8th day	Po 0th day	-	Po 20th day	-	-
Pathological results	Adenocarci noma (metastatic)	N	N	N	N	N	N	Necrotisi ng granulom atous inflamma

Table 2. Diagnosis methods and follow-up outcomes of the patients.

P: Patient, CT: Computed tomography, USG: Ultrasonography, Po: postoperative, N: normal, NA: nonavailable

4. Discussion

The most common cause of ASR in the literature is neoplastic disease (1). The results of our study indicate that, consistent with previous findings, splenic rupture associated with neoplastic disorders is quite common reason for ASR.

The major causes of ASR are classified as neoplastic disorders (including hematologic, benign, or malignant spleen-related diseases), infectious diseases, inflammatory noninfectious diseases, drugand treatment-related conditions, mechanical issues, and, lastly, cases involving a normal spleen (1). ASR can occur due to pathological stimuli affecting the spleen or in situations such as sneezing, coughing, vomiting, straining during defecation, or physical exertion (3,6).

In hematological diseases, splenomegaly due to extramedullary hematopoiesis can increase the risk of secondary ASR (7). ASR was diagnosed a patient in the 3rd trimester (mechanical disorder) with thrombocytopenic purpura (non-malignant hematological disease) in the literature. We can give this case report as an example of the etiologies that can be seen together. (8). Similarly, two of our patients had a multifactorial etiology (malignancy and hematological disease, and hematological disease and aoutoimmune disease). We also had two patients (25%) with ASR in a normal spleen, which could be considered spontaneous rupture. Although our sample size is small, the rate of spontaneous splenic rupture in our clinic was higher than reported in the literature (25% vs. 6.4%) (3).

It is well-established that ASR is associated with infectious diseases, particularly viral infections. The literature includes reports of ASR cases linked to EBV (for which physical activity is restricted for at least one month to prevent ASR) (9), CMV (10), tuberculosis (11), malaria (12), systemic salmonella infection (13), scrub typhus (14). In one patient abdominal tuberculosis was detected pathologically, and unfortunately, only three patients were evaluated with viral panels, and no active infections were detected in these cases. Through this study, we determined that ASR etiology screening in our clinic was deficient in sending viral panels, and addressed this through enhanced in-service training.

In the literature, ASR has also been reported following intra-abdominal surgery. Examples include cases of ASR secondary to bacteremia one week after an appendectomy (15), and two weeks after a laparoscopic sleeve gastrectomy (16). In our study, no ASR cases were identified in association with prior abdominal surgery.

Since ASR clinical findings can be nonspecific, imaging modalities are essential for distinguishing it from other etiologies (17). For hemodynamically unstable patients, Focused Assessment With Sonography in Trauma (FAST) USG is preferred as a rapid and non-invasive diagnostic imaging method. (7) USG is an useful first-line imaging method for detecting free intraperitoneal blood; however, its sensitivity is low while diagnosing splenic rupture (3). Multidetector CT could be primary imaging method for detecting lesions in the spleen, active bleeding, and perisplenic hemorrhage (3). Magnetic

resonance imaging, while more sensitive in detecting small intraparenchymal hemorrhages, has limited use in emergency situations due to its longer imaging time and the need for the patient to remain stable during the procedure (17).

The management of ASR, whether conservative or surgical, is related to the integrity of the splenic capsule. Patients with low-grade injuries who are hemodynamically stable can be managed conservatively (with splenic artery embolization), while surgical intervention is required for hemodynamically unstable patients (18).

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5. Conclusion

In conclusion, despite its clinically vague presentation, the diagnosis of spontaneous splenic rupture should be considered in patients with no history of trauma but who present with hypovolemic shock, acute abdomen, or abdominal pain of unknown etiology, along with a history of hematological malignancy, autoimmune diseases, or infectious diseases. Delayed diagnosis may lead to mortality.

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