

Non-traumatic torsion of an accessory spleen as a cause of acute abdomen in adults

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Abstract

Introduction: An accessory spleen (AS) is a congenital anomaly characterized by nodules of splenic tissue separate from the primary spleen, with a prevalence of 10% to 30%. Complications include torsion, infarction, cysts, and bleeding. Occasionally, AS can migrate within the abdomen, causing pain by pressing on internal organs. Torsion of AS is rare but serious, often leading to acute abdomen symptoms that may require surgical intervention.

Aim of the research: This study investigates the clinical symptoms of AS torsion and the circumstances that necessitate surgical intervention. The professional literature on AS symptoms consists only of case reports, prompting the authors to conduct a broader analysis of this condition.

Material and methods: The analysis included 24 cases of non-traumatic AS torsion in adults published between 2001 and 2024, focusing only on complete cases following the introduction of laparoscopic diagnostics. The desk research method and statistical analysis were used to assess the outcome.

Results: A total of 24 cases were identified, comprising 16 women and 8 men, aged 19 to 47 years, with a mean age of 27.1 years; half were under 24.5 years old. Abdominal pain was the most common symptom. A correct preoperative diagnosis of AS torsion was made in 8 cases. Surgical interventions included laparotomy (15 cases) and laparoscopy (9 cases), with no reported mortality.

Conclusions: AS torsion symptoms are often nonspecific, complicating diagnosis. Early recognition and surgical intervention are crucial for treatment outcomes. AS torsion should be included in differential diagnoses for the acute abdomen of unknown aetiology, particularly in young adults.

Introduction

An accessory spleen (AS) is a congenital anomaly of splenic tissue manifesting in nodules formed separately from the primary spleen [1–3]. Its prevalence varies and is estimated to be about 10–30%, depending on the study and group location, peaking in North America and Europe. In autopsies, it is over 10% [4]. In laparotomies, its prevalence is about 15% [5]. The AS can be the source of many complications, such as torsion, infarction, and cyst formation. In cases of injury, AS can also be the source of bleeding. AS can also migrate within the abdomen, causing recurrent pain by putting pressure on internal organs. An unusual location of AS can suggest a neoplasm as a differential diagnosis. However, a thorough histopathological exam can confirm the presence of splenic tissue. AS torsion is challenging to diagnose pre-operatively. Usually, when abdominal pain arises in the left hypochondriac region, the main suspicion is primary spleen pathology. It is rare for AS to be the primary source of symptoms. However, if some appropriate conditions, such as lack of fixation and ligaments, long vascular pedicle, and

migration through the abdomen occur, torsion can be possible. AS torsion is more frequent in children [6]; thus, we would like to focus on adults as a less frequent anomaly. Torsion of an AS indeed can cause an acute abdomen. Acute abdomen is a condition that requires immediate attention and treatment and is potentially life-threatening [7].

Aim of the research

The aim of the study was to analyse the clinical course of the AS in terms of the symptoms it causes that require surgical intervention.

In the professional literature, reports can be found regarding symptoms caused by AS; however, these were only case reports. This prompted the authors to conduct a broader analysis of this clinical issue.

Material and methods

The material in the study comprised 24 cases of non-traumatic torsion of the AS in adults published between 2001 and 2024 (only complete cases, not

abstracts). The material includes only articles from 2001 onward because this was the year when the first article that utilized diagnostic laparoscopy was published. The introduction of laparoscopic techniques marked a significant advancement in diagnosing and treating AS torsion. As a result, studies before 2001 did not incorporate this modern, minimally invasive approach, making them less relevant to the current understanding and management of the condition.

The desk research method was used, involving existing data described as individual cases by other researchers. The research was monographic, qualitative, and quantitative in nature, conducted in the form of a detailed description. Separate cases were obtained after searching the following Internet databases: PubMed, ClinicalKey, Academic Search Ultimate (EBSCO), BMJ Journals, Elsevier Journals, Embase, Karger, Oxford Journals, Scopus, Springer, and Wiley Online Library. Some articles were found by Google searches. All searches were performed between April and October 2024.

Each eligible case of AS torsion was entered into a case card; the data included patient age, sex, medical history, preoperative diagnosis and findings, treatment, pathology, size, and location of AS. The findings are presented in Table 1 [8–31].

Statistical analysis

The results were also subjected to statistical analysis, including the calculation of arithmetic means, standard deviations, medians, and minimum and maximum values.

Results

In total, 24 AS torsion cases were found, including 16 women and 8 men. The patients' ages ranged from 19 to 47. The mean age was 27.1 ± 7.7 , and half of the subjects were no older than 24.5. In all cases, AS was found in the abdominal or pelvic cavity.

The most common symptom reported by patients was abdominal pain, which occurred in all patients (24 cases). The side of the pain was described in 20 (83.3%) cases: in 16 cases, the pain was predominantly on the left side (66.7%), and in 4 cases, on the right side (16.7%). Tenderness was reported in 16 (66.7%) cases. A palpable mass was described by 7 (29.2%) patients. Nausea or vomiting occurred in 7 (29.2%) patients.

The correct preoperative diagnosis (torsion of AS) was made in 8 cases (33.3% of cases). The key imaging studies for establishing the diagnosis before surgery were computed tomography (CT), magnetic resonance imaging (MRI), and Doppler ultrasound.

Other indications for surgical intervention included: abdominal mass/tumour; abdominal mass with twisted vascular pedicle; acute appendicitis; acute abdominal pain; peritoneal signs; ovarian or colonic neoplasm; splenic lymphangioma; gastrointestinal

stromal tumour (GIST), tumours originating from the omentum, hemorrhagic infarction due to torsion, an organized hematoma.

In all cases of AS torsion, surgical treatment was performed with splenectomy of AS by laparotomy (15 cases; 62.5% of cases) and by laparoscopy (9 cases; 37.5% of cases).

No mortality has been reported.

Discussion

The accessory spleen usually gains clinical significance in cases of total splenectomies (including primary and accessory spleens) for haematological diseases or immune thrombocytopenic purpura (ITP). Incomplete resection of splenic tissue may result in the need for reoperation [32, 33]. AS also plays a crucial role when detected during radiological procedures. Accessory spleens may be detected incidentally and must be carefully distinguished from conditions such as splenosis, neoplasms, or lymphadenopathy. When the pain in the abdomen occurs, AS can also be a possible cause. Usually, AS is asymptomatic. In certain conditions, such as torsion, rupture, enlargement of AS, and haemorrhage, AS may produce symptoms [34]. Usually, when AS becomes symptomatic, the symptoms are not specific. They may be associated with location, such as pain in the left upper quadrant of the abdomen. Still, AS can also wander through the abdomen, for example, mimicking acute appendicitis [35]. The differential diagnosis of AS torsion should include a broad spectrum of abdominal diseases. The symptoms of AS torsion (abdominal pain, tenderness, palpable mass, nausea, vomiting) are very similar to those described in primary spleen torsion [36]. The complaints reported by the patient may also resemble subserosal torsion of uterine myoma [37, 38]. The most prevalent locations of AS are the splenic hilum, the tail of the pancreas, gastrosplenic and lienorenal ligaments, the greater omentum, the mesentery [34, 39]. AS can also be located in the pelvis [40] and the thorax [41].

Torsion of AS is a rare condition but can be the cause of acute abdomen. As it manifests in a non-specific manner, just like the torsion of a primary spleen, it often requires imaging diagnostics to be diagnosed [42]. Depending on the availability and stability of the patient, it is usually determined through USG, CT, or MRI. The condition manifests as a lack of flow through Doppler USG. If the patient is stable, 99mTc-denatured RBC scans can be helpful in identifying the AS [3, 43]. However, in cases of hemodynamical instability caused by internal bleeding, the diagnosis is made during emergency surgery. The laparoscopic/robotic/minimally invasive surgery seems to be a proper approach. However, it may not be attainable in cases of extensive abdominal masses. Thus, laparotomy is always an applicable approach.

Table 1. All the analysed cases

No.	First author of case, year, citation	Age	Sex	Medical history	Preoperative diagnosis/preoperative findings	Treatment	Pathology	Dimensions of AS [cm]	Localization of AS
1	d'Halluin, 2001 [8]	26	F	Acute and severe abdominopelvic syndrome, exquisite tenderness in the Douglas pouch	Operated due to acute pain; a small amount of fluid on the pelvic USG	Emergency laparoscopy	Torsion and infarction of an AS	4	In the pelvis, located on the posterior aspect of the uterine isthmus
2	Fujita, 2002 [9]	23	M	Intermittent left-sided abdominal pain and persisting fever of about 37°C	Abdominal tumour on USG, CT, MRI	Laparotomy	A solid tumour connected to a twisted twice pedicle about 5 cm in length, believed to be a branch of the left gastroepiploic artery and vein; the tumour was composed of splenic tissue with extensive necrosis, fibrosis, and granulation tissue; the tumour was diagnosed as an AS that had undergone infarction due to torsion	8.0 × 7.2 × 4.8	Within the greater omentum
3	Grinbaum, 2005 [10]	21	F	2-day history of increasing, intense, left upper abdominal and left flank pain; left upper quadrant abdominal mass, tenderness with voluntary guarding	Abdominal mass with twisted vascular pedicle on CT; no blood flow on Doppler USG	Laparoscopy	Histologic examination was consistent with infarcted splenic tissue.	7.5 × 6 × 7	Left mid-abdomen
4	Jung, 2005 [11]	25	F	Acute onset of low abdominal pain and a large palpable mass at the left lower quadrant of the abdomen	Peritoneal mass with twisted vascular pedicle on CT; avascular on Doppler USG	Laparotomy	An AS with hemorrhagic infarction. A long and mobile vascular pedicle arising from the splenic hilum (arrows) was torsed in a clockwise direction 10 times	4	Near the left lower quadrant of the abdomen
5	Corsi, 2007 [12]	37	F	Left upper abdominal pain spreading to the homolateral lumbar region, clinical exacerbation towards an acute abdomen	Peritoneal tumour on USG, CT	Emergency surgery due to the patient's worsening symptoms	Torsion of AS	9.3 × 6.5 × 9	Left mid-abdomen

Table 1. Cont.

No.	First author of case, year, citation	Age	Sex	Medical history	Preoperative diagnosis/preoperative findings	Treatment	Pathology	Dimensions of AS [cm]	Localization of AS
6	Nishigori, 2007 [13]	32	M	Severe left abdominal pain, marked tenderness, progressive anemia	A splenic tumour (abdominal mass with the splenic artery branches serving as the tumour vessels on CT)	Emergency laparotomy with AS splenectomy	Severe congestion in the splenic tissue, red thrombi in the arteries and veins; no malignant findings. The vascular pedicle, branching from the splenic artery and vein, was twisted 720° clockwise. Intraperitoneal haemorrhage due to a ruptured splenic tumour	7 × 7 × 4	Pancreas tail
7	Kim, 2009 [14]	24	M	3-day history of epigastric pain that was accompanied by nausea and vomiting. Blood tests revealed an elevated white cell count and a mild elevation of erythrocyte sedimentation rate and CRP	Abdominal mass with whirled tubular structure on USG, CT; no blood flow on Doppler USG	Laparotomy with AS resection	Violet, oval tumour under the omentum with a twisted vascular pedicle attached to the splenic pedicle; pathologic examination confirmed torsion and hemorrhagic infarction of an AS	8	The left upper abdomen
8	Takemoto, 2009 [15]	34	F	Pain in the left upper abdomen and flank; palpable 10 cm mass in the same region with tenderness	Gastrointestinal stromal tumour (GIST), tumours originating from the omentum, hemorrhagic infarction due to torsion, an organized hematoma (suggestion of vascular constriction due to torsion based on CT; suggestion of internal haemorrhage based on MRI)	Exploratory laparotomy	A bluish congested mass with a 360° twisted pedicle connected to the splenic artery and vein. Torsion of an AS was diagnosed	10	The left upper quadrant of the abdomen
9	Morita, 2012 [16]	37	F	Sudden left upper quadrant abdominal pain, fever for several days, tenderness in the left upper abdomen and periumbilical region with a palpable soft mass	Torsion of an AS (primary diagnosis), duplication cyst or epidermoid cyst based on CT and MRI findings	Emergency laparotomy	Although the tumour had undergone infarction, it had a structure resembling spleen tissue, strongly suggesting an AS. Despite the extensive necrosis, silver staining revealed splenic sinus-like structures	6.7 × 4.3	Near the tail of the pancreas

Table 1. Cont.

No.	First author of case, year, citation	Age	Sex	Medical history	Preoperative diagnosis/preoperative findings	Treatment	Pathology	Dimensions of AS [cm]	Localization of AS
10	Sadrol, 2013 [17]	22	F	Acute onset of left-flank pain upon waking in the morning. The pain was sharp and non-radiating. There was nausea but no vomiting. Pertinent past medical history included gastric bypass surgery 18 months before	Two ASs, torsion of a larger AS with infarction based on CT	Emergency surgery due to persistent pain	AS was discovered to be infarcted with fibrosis	Smaller one 0.9; larger one 1.9 × 1.9 × 2.7	Medial and anterior to the lower pole of the spleen
11	Bard, 2013 [18]	20	F	5-day history of left-sided abdominal pain accompanied by loss of appetite and a single vomit; a soft abdomen and a tender palpable mass in the left side of her abdomen. Diagnosis of asymptomatic AS 10 years earlier	A huge non-enhancing mass consistent with an ischemic giant AS with a twisted vascular pedicle on CT angiography	Small midline laparotomy	Ischemic accessory, huge spleen on a long twisted pedicle	17 cm in the largest diameter	The lower pole of a small native spleen in its normal anatomical location
12	Kee, 2014 [19]	19	F	3-day history of left-upper quadrant pain that became slowly aggravated; left-side abdominal tenderness, most markedly in the left upper quadrant, decreased bowel sounds, temperature 37.7°C	Torsion of an AS on CT	Emergency exploratory laparoscopy with accessory splenectomy	Large, rounded violet mass with a twisted vascular pedicle located anterior to the normal spleen; acute hemorrhagic infarction of AS	Unknown	Left upper abdomen, located anterior to the normal spleen
13	Ozeki, 2015 [20]	31	F	Intense left abdominal pain, pyrexia (38.5°C); a left upper quadrant abdominal tenderness with voluntary guarding; an elevated level of serum C-reactive protein	Torsion of AS on CT	Single-port laparoscopy	AS with hemorrhagic infarction and torsion of the stalk; 180° torsion around a long vascular pedicle along the left side of the greater omentum	3 × 3 × 3.5	Greater omentum
14	Sugiura, 2015 [21]	23	F	Increasing abdominal pain; abdominal distension, tenderness, guarding over the entire lower abdomen, and rebound tenderness in the right lower quadrant, temperature 37.6°C	Acute appendicitis/abdominal tumour on USG, CT, MRI	Emergency laparotomy	The tumour with two twisted vascular pedicles was in the pelvis and excised. On macroscopic examination, the tumour was 14 cm × 11 cm and red inside. Microscopically, it contained congestive splenic tissue. The final diagnosis was torsion of an AS	14 × 11	Left upper quadrant of abdomen

Table 1. Cont.

No.	First author of case, year, citation	Age	Sex	Medical history	Preoperative diagnosis/preoperative findings	Treatment	Pathology	Dimensions of AS [cm]	Localization of AS
15	Samir, 2017 [22]	27	F	4-day history of left-lumbar region pain that slowly aggravated and became continuous; left-side abdominal tenderness, most markedly in the left lower quadrant, decreased bowel sounds, palpable vague firm mass in the left lumbar region; temperature 37.7°C; 14th week of pregnancy	Torsion of an AS on MRI	Exploratory laparotomy with accessory splenectomy	A large, rounded violet mass with a twisted vascular pedicle located in the left lumbar region with a small bowel and mesentery adhered to it	Unknown	The lumbar region
16	Termos, 2017 [23]	27	F	Few hours' history of severe diffuse abdominal pain, mainly in the left upper quadrant (sudden in onset, aching, radiating to the left intra-scapular area and left shoulder), associated with nausea and 3 episodes of non-bilious and non-bloody vomiting; marked upper abdominal tenderness mainly over the epigastric area and left hypochondrium with voluntary guarding; anemia; temperature of 37.5°C	Surgical intervention due to radiological findings (avascular mass on CT and Doppler USG) and in view of the intolerable persisting abdominal pain	Laparotomy (small upper midline incision), splenectomy of AS	Advanced ischemic changes with haemorrhage of splenic parenchyma; findings were consistent with splenic infarction	13 × 6 × 3.2	Left upper quadrant
17	Watanabe, 2017 [24]	22	M	Left-sided abdominal pain; tenderness to the left side	Torsion of AS on CT	Emergency laparoscopic surgery	Histopathologically, the dark-red mass was diagnosed as torsion of an AS	1	Greater omentum
18	Bowles, 2021 [25]	43	F	2-day history of abdominal pain; the pain began suddenly whilst at rest and was located centrally within the abdomen with later migration to the left iliac fossa; nausea; tenderness in the left lower quadrant with localized peritonism	Malignancy, infection (believed less likely), ovarian or colonic neoplasm with peritoneal nodularity and a large soft tissue deposit based on mass with a feeding vessel on CT	Emergency diagnostic laparoscopy	Well-encapsulated mass with marked haemorrhage but with residual architecture consistent with splenic tissue. The other vessels were congested with blood clots. The features were in keeping with the AS with infarct	6.5 × 3.1 × 2.7	Left lower quadrant of the abdomen

Table 1. Cont.

No.	First author of case, year, citation	Age	Sex	Medical history	Preoperative diagnosis/preoperative findings	Treatment	Pathology	Dimensions of AS [cm]	Localization of AS
19	Diamond, 2022 [26]	20	M	A week-long history of persistent abdominal pain and fever, worsening of symptoms	Large AS with volvulus resulting in an infarction or injury on CT	Laparoscopic splenectomy	Torsed AS with haemorrhage, vascular congestion, and infarction; fat necrosis and inflammation	14.0 × 7.5 × 4.8	Left upper abdomen, within the mesentery
20	Sayyadi, 2022 [27]	23	M	Abdominal pain in the epigastric region, which was migrated to the right lower quadrant after 5 h of onset and intensified, a lack of appetite, nausea, and five episodes of non-bilious, non-bloody emesis, an increased temperature (38.1°C); tenderness and rebound tenderness in the right lower quadrant	Acute appendicitis	Urgent laparotomy, appendectomy with AS splenectomy	Intraoperative finding: an inflamed appendix with a solid lesion suspected of AS in the mesoappendix. Pathology report: acute appendicitis, the solid mass findings were indicative of normal spleen tissue	2	Mesoappendix
21	Montero Verdecia, 2022 [28]	21	M	Abdominal pain that began 1 day before in the mesogastrium and then moved to the right lower quadrant, which intensified progressively, was constant, without radiation, did not relieve with pain relievers, accompanied by anorexia, nausea, increased with sudden movements; fever (38.5°C); pain on superficial and deep palpation in the right lower quadrant, with muscular defence and peritoneal reaction	Surgery due to peritoneal signs	Emergency laparoscopy	An ischemic AS with torsion of the vascular pedicle; histology examination: splenic tissue with areas of infarction	4	At 60 cm from the ileocecal valve next to the mesenteric border of the jejunal loop
22	Emir, 2023 [29]	47	F	Nausea, vomiting, abdominal pain more apparent in the left upper quadrant, tenderness, defence, rebound	Abdominal mass on USG, CT	Exploratory laparotomy with accessory splenectomy	Exploration detected a bluish-coloured, congested, 720°-rotated pedicled, mobile mass located in the left upper quadrant and surrounded by the omentum -ectopic splenic tissue showing signs of hemorrhagic infarction	12 × 7 × 4.8	Adjacent to the left kidney

Table 1. Cont.

No.	First author of case, year, citation	Age	Sex	Medical history	Preoperative diagnosis/preoperative findings	Treatment	Pathology	Dimensions of AS [cm]	Localization of AS
23	Haidari, 2023 [30]	27	M	Persistent left hypochondrial pain of 3 days' duration	Splenic lymphangioma based on CT	Exploratory laparotomy	Splenic parenchyma consisting of red and white pulp with diffuse haemorrhage, dilated blood vessels, and areas of infarction that lead to the diagnosis of an AS with torsion	15 × 7	Left upper quadrant of the abdomen
24	Locurto, 2024 [31]	19	F	Sudden upper abdominal pain, hypotension; abdominal tenderness mainly in the right flank and upper abdomen with mild peritoneal signs of rebound and guarding, a palpable mass in the periumbilical area, remarkable abdominal bloating, torpid peristalsis in auscultation; 2 months earlier, diagnosed with membranous glomerulonephritis and signs of nephrotic syndrome	Extremely mobile and bleeding abdominal mass changing its position between CT and MRI scans	Emergency exploratory laparoscopy, splenectomy of AS	A pedunculated nodular mass, brownish-grey coloured, with own capsule; splenic parenchyma type architecture with thickened fibrous capsule and congested dilated vessels with a sinusoid appearance, follicular formations with diffuse central reactive hyperplasia phenomena and minute arterioles with thickened walls and sometimes fibrotic (penicillary-like arteries); [...] chronic ischemic phenomena due the probable torsion of the peduncle [...]	6.9 × 4.8 × 5.3	Variable position: left abdomen (in correspondence of the mesentery, close to the left rectus abdominis muscle) on CT; right upper abdomen (in front of the right kidney, close to the right rectus abdominis muscle, with coarse vascular pedicle) on MRI; close to the transverse mesocolon during the surgery

AS – accessory spleen, USG – ultrasonography, CT – computed tomography, MRI – magnetic resonance imaging, CRP – C-reactive protein, GIST – gastrointestinal stromal tumour.

Conclusions

AS torsion is a rare condition. Almost all of the cases involved individuals aged up to 40. AS torsion should always be considered a possible cause of acute abdomen of unknown aetiology, especially in young adults, who were dominant in our study. The symptoms of AS torsion do not differ from those of the primary spleen torsion. The AS may be misdiagnosed as an abdominal neoplasm of nearby affected organs, especially when it is asymptomatic. Nevertheless, if acute abdomen symptoms occur, AS should always be excised. Preoperative identification of an AS or the absence of a primary spleen facilitates patient management and shortens the duration of surgery.

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Ethical approval

Not applicable.

Conflict of interest

The authors declare no conflict of interest.

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