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Review

The prevalence and morphometry of an accessory spleen: A meta-analysis and systematic review of 22,487 patients

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HIGHLIGHTS

- Accessory spleen is a common variant, present in almost 15% of the population.
- It is relatively more common in patients with immune thrombocytopenia.
- A quarter of patients with accessory spleen have more than one accessory spleen.
- Identification of accessory spleen in immune thrombocytopenia patients is essential.

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ABSTRACT

Background: An accessory spleen (AS) is a lobule of splenic tissue found in ectopic locations. Identification of AS is particularly important in patients with immune thrombocytopenia (ITP) requiring splenectomy as unrecognized AS can later cause refractory symptoms. The AS can also be a source of significant intraabdominal hemorrhage. The aim of this meta-analysis was to systematically analyze the data on the prevalence, number, location, and morphometry of AS.

Materials and methods: An extensive search of the major electronic databases was conducted to identify all studies that reported relevant data on the AS. No date or language restrictions were applied. Data on the study type, the prevalence of AS, location, morphometry and number of AS per patient were extracted from the eligible studies and pooled into a meta-analysis.

Results: A total of 81 studies (n = 22,487 subjects) were included into the quantitative analysis. The overall pooled prevalence of AS was 14.5% (95%CI: 12.4–16.7), while the pooled prevalence of AS in ITP patients was 16.7% (95%CI: 12.1–21.7). The majority of accessory spleens were located in the splenic hilum (62.1% [95%CI:51.5–76.3]). Moreover, 26% of ITP patients with an AS have more than one.

Conclusions: The findings of this study provide an evidence-based foundation of anatomical knowledge about the AS. Surgeons should take particular caution in identifying an AS, as unnoticed AS during splenectomy can lead to recurrence of hematological diseases or can be a potential source of bleeding in the future.

1. Introduction

An accessory spleen (AS) or splenunculus is a lobule of splenic tissue observed in ectopic locations [1] (Fig. 1). Identification of an AS is of crucial importance in various clinical situations, including abdominal trauma, as well as in hematological, immunological and lymphoproliferative disorders. Failure to recognize and remove the

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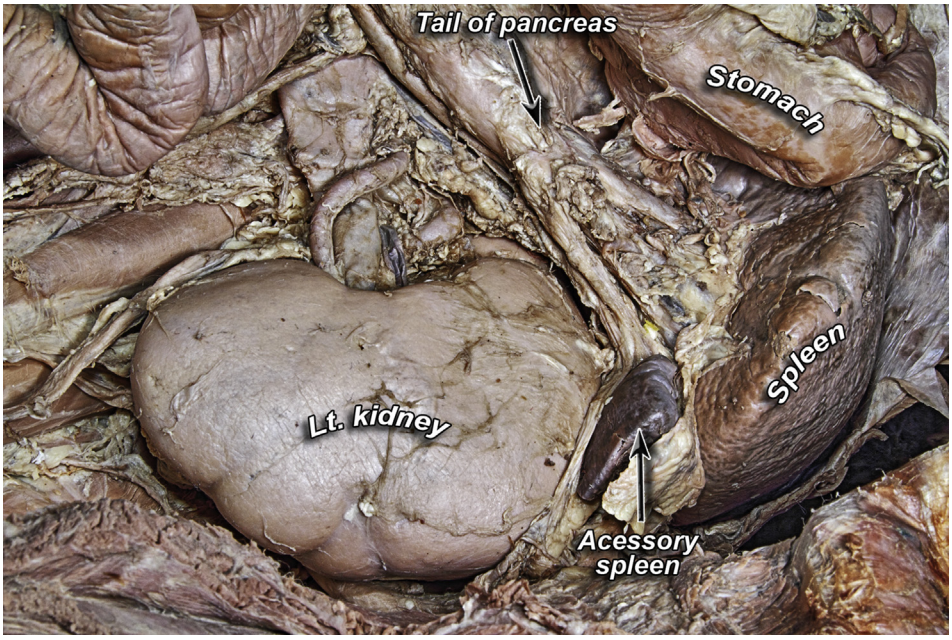

Fig. 1. An accessory spleen in the splenorenal ligament.

AS could cause refractory or recurrent disease in patients with immune thrombocytopenia (ITP, previously termed idiopathic thrombocytopenic purpura) requiring splenectomy due to insufficient response to medical therapy with glucocorticoids or intravenous immunoglobulins [2]. Also, splenic tissue is highly vascularized, and spontaneous or traumatic rupture of an AS could cause significant intraabdominal hemorrhage [3]. Moreover, the AS might be misdiagnosed as enlarged lymph node or as a tumor, especially when localized in or near the pancreas.

An AS forms when the splenic tissue fails to fuse properly in the dorsal mesogastrium during embryologic development [4]. The AS is most commonly located in the hilar region of the spleen, the mesentery, along the splenic vessels or in the greater omentum, but other various locations have been reported [5]. Accessory spleens are usually single, but cases of more than three in a single patient have been reported in the literature [5–12]. Morphologically, AS forms uniform and round or oval lobules. While many cadaveric, imaging, and intraoperative studies have investigated AS, the reported prevalence of this structure is inconclusive and ranges from less than 1% [13] to 76% [14]. Also, geographical differences in its prevalence have been shown [15–17], but poorly studied. The AS can be detected by ultrasonography (USG), contrast-enhanced USG, computed tomography (CT) and magnetic resonance imaging (MRI) [18]. Scintigraphy with technetium sulfur colloid can provide a definitive identification of the AS [19].

Due to its clinical significance, detailed knowledge on the anatomy and prevalence of this variation is essential for physicians, surgeons, and radiologists. However, to date, the literature lacks conclusive data on the epidemiologic and anatomic characteristics of the AS. The aim of this meta-analysis was to systematically analyze the literature and provide evidence-based data on the prevalence, number, location, and morphometry of accessory spleens.

2. Methods

2.1. Search strategy

An extensive search of the major electronic databases, including PubMed, Embase, ScienceDirect, SciELO, BIOSIS, and Web of Science, was conducted up to March 2017 in order to identify all studies reporting relevant data about the AS. The following search terms were employed: accessory spleen OR supernumerary spleen OR splenunculus OR splenunculi OR splenule OR splenules. No date or language restrictions were applied during the search. An additional search through the references of the identified studies was conducted. The protocol for this study strictly adhered to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (*PRISMA*) guidelines [Supplement 1].

2.2. Eligibility assessment

The eligibility assessment for the inclusion into this meta-analysis was conducted by three independent reviewers (JV, BS, DT). Peer-reviewed cadaveric, imaging or intraoperative studies that reported the prevalence and anatomical data of AS were included into the meta-analysis. Conference abstracts, letters to editor, reviews and studies providing incomplete data were excluded. Studies in languages other than English were translated by medical professionals fluent in both English and the original language of the article and further evaluated. In case of any disagreement between the reviewers during the eligibility assessment, a decision was made through a consensus process among the review team.

2.3. Data extraction

Data extraction was conducted by three independent reviewers (JV, BS, DT). Data on the study type, the prevalence of AS and number of AS per patient were extracted from the eligible studies. The patients were also divided in relation to their health status into ITP group and healthy patients (defined as trauma patients - without known hematological, neoplastic or other intraabdominal pathology, as well as patients reported as healthy controls). The authors of the original articles were contacted, when possible, if any discrepancies in the study data was observed or further details were needed.