

Case Report

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Exceptional Primary Squamous Cell Carcinoma of the Spleen: A New Case Report and Literature Review

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ABSTRACT

Squamous cell carcinoma (SCC) is a malignant epithelial neoplasm showing squamous differentiation. Primary splenic SCC is exceedingly rare, with only one previously reported case in the literature to date. Preoperative diagnosis is challenging, because of the lack of specific imaging features. Surgical management is indicated for diagnostic and therapeutic purposes. Definitive diagnosis requires histopathological analysis. Herein, we describe the second case of splenic squamous cell carcinoma, in a 60 yo women, with fatal outcome, to raise awareness of this entity and to highlight the diagnostic difficulties in such a presentation.

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Introduction

Squamous cell carcinoma (SCC) is a malignant epithelial neoplasm showing squamous differentiation. It is frequently reported in the head and neck region as well as the skin [1]. Primary splenic SCC is exceedingly rare, with only one previously reported case in the literature to date [2]. Preoperative diagnosis is challenging; the definitive diagnosis requires histopathological analysis [3]. Herein, we describe the second case of splenic squamous cell carcinoma, in a 60 yo women, with fatal outcome, to highlight the diagnostic difficulties in such a presentation.

Case Presentation

60 Yo women presented to department of surgery with a 3 months history of pain and swelling in the left hypochondrium. She had a history of type 2 diabetes and Biermer' disease without any previous gynecological complaints. Physical examination revealed a palpable mass and tenderness of the left hypochondrium. The general condition was good, with stable vital signs. There were neither lymph nodes nor cutaneous lesions. Blood analysis showed a slight anemia with hemoglobin at 10 g/dL. Tumor markers (carcinoembryonic antigen, CA 19-9, alpha-fetoprotein) and other parameters were within normal ranges. Computed tomography revealed a huge tissular mass in the splenic hilum with heterogeneous enhancement and central necrosis measuring :18 x 14 x 12 cm [Figure 1], infiltrating the stomach, the pancreas and the splenic artery, with multiple peritoneal lymphadenopathies. These findings were suggestive of splenic lymphoma with nodal involvement. Surgical exploration found splenic tumor infiltrating pancreatic tail and the transvers mesocolon. The biopsy has accidentally led to a discharge of a necroticopurulent liquid. The sample was submitted for intra-operative frozen section consultation, which showed poorly differentiated

carcinoma. Since the tumor was locally advanced, multiple biopsies were performed for further pathological assessment. Histology revealed a carcinomatous proliferation consisting of squamoid cells with abundant cytoplasm, intercellular bridges, pleomorphic nuclei, prominent nucleoli and mitotic figures. These cells were arranged in confluent nodules, centered by keratinization. The stroma showed abundant lymphocytes [Figure 2]. No splenic parenchyma was found in all fragments. Due to the scarcity of splenic SCC, immunohistochemistry was performed to rule out a non squamous associated component. It showed diffuse positivity for p40 [Figure 3]. As p16 is not available in our institution, human papillomavirus (HPV) associated SCC could not be ruled out.

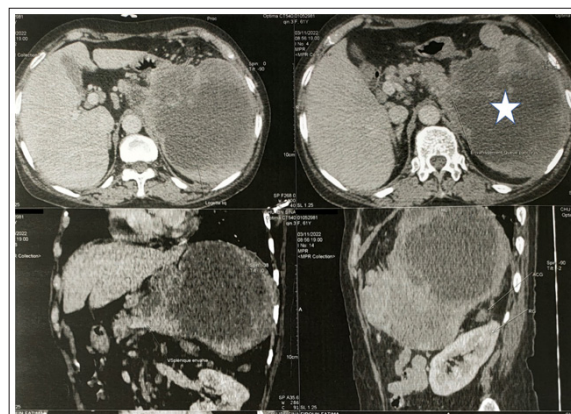


Figure 1: Abdominal computed tomography (CT) scan showed a 18 x 14 x 12 cm tissular heterogeneous mass, with irregular borders, located in the splenic hilum with extensive necrosis (white star)

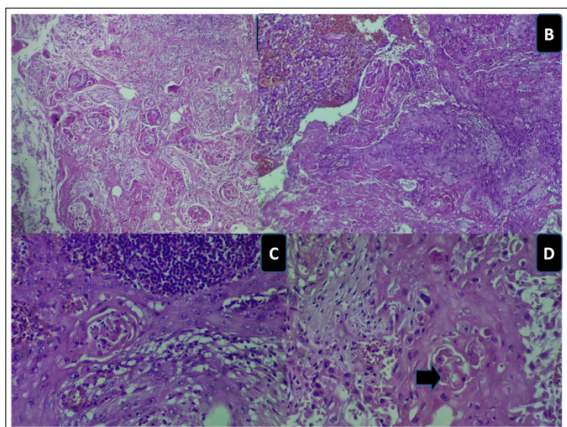


Figure 2: Splenic squamous cell carcinoma histological features: (A) hematoxylin and eosin, $\times 100$ (B) hematoxylin and eosin, $\times 200$ (C) And (D) hematoxylin and eosin, $\times 400$ (black arrow: keratin pearls)

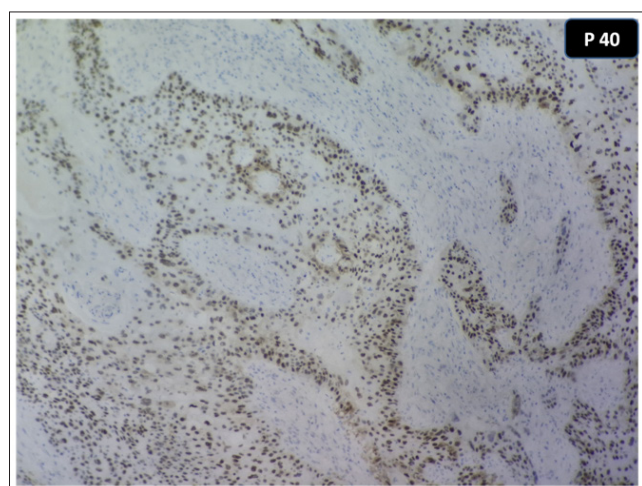


Figure 3: Splenic squamous carcinoma immunohistochemistry: the carcinomatous cells diffusely positive for p40

After surgery, the patient had further investigations to delineate any extrasplenic involvement including: radiological assessment (pelvic MRI, thoracic Ct scan) and gynecological examination (colposcopy and biopsy). They both failed to find a primitive squamous cell carcinoma elsewhere. In the absence of extrasplenic localization, the definitive diagnosis was primary splenic locally advanced squamous cell carcinoma.

Since the patient was not eligible for radical surgery, she was discharged with an uneventful post operative course, and then addressed to oncology department for further management. She died 2 months after the surgery; the cause of death remained unknown as the patient died at home.

Discussion

Splenic tumours are rare, representing a wide range of different neoplasms: benign entities, and malignant primary or secondary tumors. Given the composition of splenic parenchyma (lymphoid and vascular elements), splenic lesions can be divided into:

- Lymphoid
- Non lymphoid, among which vascular tumors are the most common; including: angiosarcoma, hemangiomas, lymphangiomas, and hemangioendotheliomas [3].

Splenic SCC are exceedingly rare, represented by metastasis, from primary lesions of: lungs, stomach, pancreas, liver, colon and oesophagus [4, 5]. To the best of our knowledge, this is the second case of primary splenic SCC reported in the English literature to date. In fact, the first case was reported by Jiang Z. et al. [2]. The pathogenesis of splenic SCC remains unclear, it can be secondary to a squamous metaplasia, or a metastatic disease of SCC with a false negative staging [5]. In our patient, the first hypothesis is more plausible, as a metastatic SCC was ruled out.

The diagnosis requires a histological proof. SCC is a malignant epithelial tumor composed of cells showing varying degrees of:

- morphologic squamous differentiation: (intercellular bridges and or keratinization);
- and /or phenotypic squamous differentiation (expression of squamous markers p40, p63, or CK5/6).

SCC is graded as well, moderately, or poorly differentiated, according to the degree of differentiation, pleomorphism and mitotic activity [1]. It might be a part of a composite or collision tumor defined as “Tumors with 2 different histological morphologies” Thus, extensive sampling is mandatory to rule out another associated component [7]. In our case, multiple peroperative biopsies excluded a non squamous component.

The preoperative diagnosis of splenic tumors is challenging, as these tumors are usually asymptomatic [3]. Cross-section imaging is an important tool for detecting and characterizing splenic tumors ; especially FDG PET scan. It shows, an up to 100% sensitivity and specificity in distinguishing benign splenic lesions from malignant ones [8]. In our patient, the diagnosis of malignancy was straight forward as the tumor was infiltrating adjacent organs, however, metastatic disease could not be excluded. In fact metastasis might present as large masses with cystic centers and heterogeneous enhancement [8].

Given the lack of specific imaging features in splenic solid malignancies, percutaneous biopsy could be an important diagnostic tool. Nevertheless, when considering the risk-benefit ratio (bleeding and capsule's rupture), surgery is indicated for diagnostic purposes. In fact, surgical management of splenic tumors represents a diagnostic tool and a therapeutic option as well. In the series of Gonzalez et al. splenectomy was performed in all patients, without preoperative biopsy [3].

Surgical management include open and minimally invasive surgery. When technically feasible, the laparoscopic approach is indicated to reduce postoperative complications. Due to the scarcity of splenic malignancies, the indications for surgical treatment is challenging. Surgical resection might be indicated for primary as well as splenic oligometastasis from an oncological point of view [9].

Conclusion

This case reports the differential diagnosis of splenic solid tumors and highlights the challenging preoperative diagnosis. Percutaneous biopsy may not be possible, and doesn't affect the treatment or the surgical outcome. Therefore, surgical management is necessary for a definitive diagnosis. Finally, it is worth mentioning that a primary pancreatic SCC cannot be ruled out given the advanced stage of the tumor, therefore this case report still rare because of the scarcity of this presentation as well.

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- **Author's Contributions:** All authors read and approved the final manuscript.

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