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Non Traumatic Rupture of Splenic Metastases as the First Presentation of Clinically Occult Disseminated Primary Lung Cancer

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Key Learning Points

- Though ruptured aortic aneurysm is one of the commonest causes of sudden hypovolemic shock in an adult male, an urgent abdominal CT angiography is recommended for confirmation of diagnosis as it may occasionally reveal an intact aorta with other unexpected causative factors such as an unsuspected splenic rupture.
- Non traumatic splenic rupture is rare and is usually associated with a 'pathological' spleen with underlying infectious or neo plastic lesions which are usually primary hemopoietic in nature with secondary metastases being relatively uncommon.
- Metastases to the spleen from visceral organs is extremely infrequent (<5%) with breast, lung, pancreas, ovary, melanoma and choriocarcinoma often being attributed as their primary source.
- Though lung cancer is a commonly diagnosed cancer in Canada with many being diagnosed at an advanced stage, non-traumatic rupture of splenic metastases is an extremely rare first time clinical presentation of primary occult disseminated lung cancer.

1. Abstract

In Canada, lung cancer is one of the most commonly diagnosed cancers and is the leading cause of cancer related death. Unfortunately, more than half of all lung cancers present at an advanced stage and are often metastatic at diagnosis. The common sites of metastases include the brain, bone, liver, adrenals, opposite lung and distant lymph nodes. The spleen, a hemopoietic organ is an extremely uncommon site of metastases. In the setting of lung cancer, splenic metastases occurs in the context of disseminated disease, or uncommonly presents as a solitary metastases that is often detected on radiological image surveillance. Most of these scenarios occur in the background of known lung cancer disease.

In this case report, we share a case of non-traumatic rupture of splenic metastases whose clinical presentation mimicked a ruptured abdominal aneurysm. The presentation of non-traumatic rupture of the splenic metastases arising from an occult disseminated primary lung cancer as the first clinical encounter is extremely rare and to our knowledge has not been previously reported. Increased clinical awareness of this extremely uncommon clinical presentation of an underlying common disease is important as rapid diagnosis can significantly reduce major morbidity and mortality.

2. Case Presentation

A 63-year-old male presented to the community hospital with a one hour history of sudden onset of central abdominal pain, nausea, vomiting and diarrhea accompanied by shortness of breath. On examination, his abdomen was mildly distended with signs of peritonism. His past medical history included a remote repair of an inguinal hernia and was otherwise unremarkable. While being transferred to a larger referral center for further workup and investigations, he unfortunately went into cardiac arrest requiring cardiopulmonary resuscitation. He was intubated on arrival at the emergency department as he was hemodynamically unstable and in acute hypovolemic shock. Vascular surgery was consulted for a suspected ruptured abdominal aortic aneurysm.

After initial assessment and resuscitation, the patient underwent urgent Computed Tomography Angiography (CTA) that showed no evidence of a ruptured abdominal aortic aneurysm. The CT scan however showed evidence of a splenic rupture as seen in (Figure 1) that was associated with a hilar mass(Figure 2) which was suspicious for primary lung malignancy with metastatic disease to the liver (Figure 3) and adrenals.

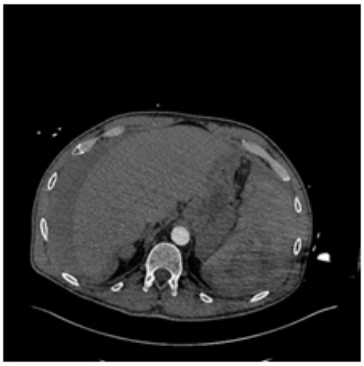


Figure 1: CT Scan shows normal caliber aorta with no evidence of aneurysm. In the left upper quadrant the spleen is not well demarcated and there is a large amount of heterogeneous suspected clot.

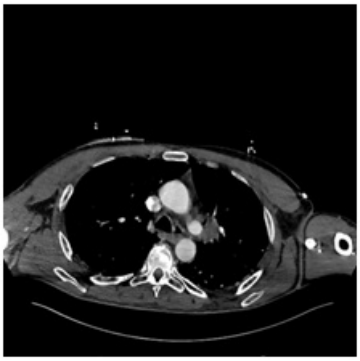


Figure 2: CT Scan shows a lobular left suprahilar mass measuring 3.1x2.7 accompanied by a lymphnode suspicious for lung malignancy.

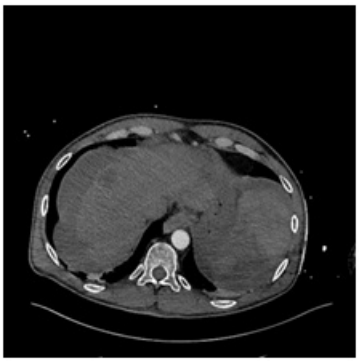


Figure 3: CT scan shows suspicious multiple hypodense lesions in the liver in keeping with metastatic lesions from lung primary.

Emergency laparotomy confirmed a large volume of blood within the peritoneal cavity with an intact aorta and continual ooze from the left upper quadrant. Having achieved hemostasis, aruptured spleen with an intrasplenic solid mass was identified leading to an emergency splenectomy. Additionally, there were several intra abdominal lesions in both lobes of the liver as well as intrathoracic findings suspicious for a lung primary. Although his surgery was uneventful, the patient failed to respond to continued resuscitation and eventually went into intractable shock leading to his death within the next forty-eight hours.

2.1. Pathology

2.1.1. Gross

A spleen measuring 7.7x5.2x2.5cm and weighing 66.2 gm with a partially stripped capsule and dark red brown diffusely nodular surface was received in the pathology laboratory. Sectioning of the spleen revealed the presence of a single fairly well delineated tan white mass measuring 2.3 x 2.2 x 3.3 cm with necrotic and hemorrhagic areas (Figure 4).

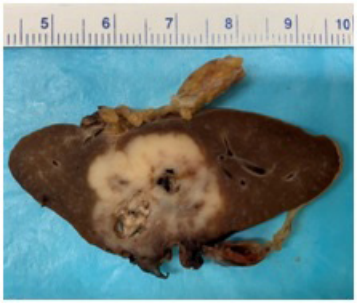
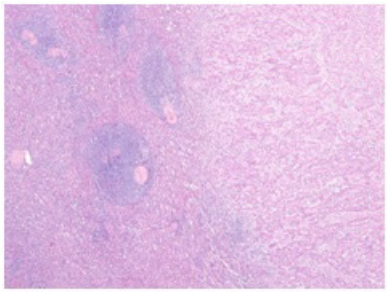


Figure 4: Cut section of the spleen shows a fairly well circumscribed tan necrotic mass with hemorrhage measuring 2.3x2.2x3.3cm.

2.1.2. Histopathology

Representative sections confirmed the presence of a non-splenic/ non hematopoietic mass that had an ill-defined border and was composed of neoplastic cells that had a vague glandular formation in keeping with a metastatic adenocarcinoma as seen in (Figures 5A and 5B).

5A



5B

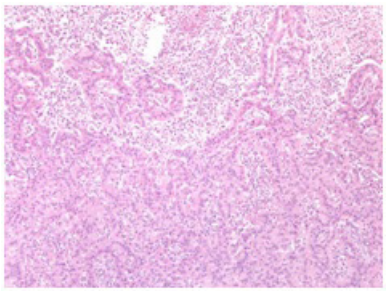


Figure 5: Photomicrographs of haematoxylin and eosin stained slides at medium power shows a fairly well demarcated neoplastic lesion composed of 'nonsplenic' tissue [5A]. At high power, the neoplastic cells are seen to form tubules and glands in keeping with a metastatic adenocarcinoma [5B].

Immunohistochemical staining with adequate positive and negative controls confirmed the presence of an epithelial malignancy with diffuse strong expression of cytokeratin 7 (Figure 6A) and no expression of BCL2/ CD20 (Figure 6B) that was expressed as expected in the native white pulp of the surrounding splenic parenchyma. There was no expression of CK20 or CDX2(Colorectal markers)in the lesional cells. Lineage specific immunohistochemical markers confirmed the tumor to be lung in origin with diffuse strong nuclear expression of TTF1 (Figure 6C) and co-expression of Napsin (Figure 6D).

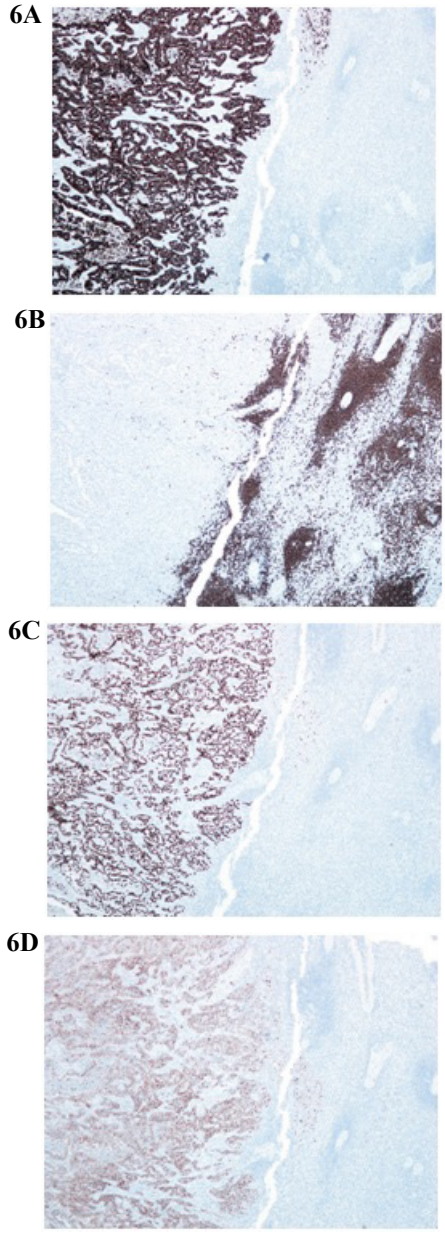


Figure 6: Photomicrographs of immunohistochemical stained slides with adequate positive and negative controls show the neoplastic cells to strongly express Cytokeratin 7 [6A] and are negative to native splenic cells that are stained positive with BCL2/CD20 in 6B.
Lineage specific immunohistochemical markers show strong positive nuclear staining with TTF1 [6C] supporting lung primary accompanied by diffuse expression of NAPSIN A [6D] in keeping with bronchogenic adenocarcinoma histological subtype.

3. Discussion

Non traumatic splenic rupture is uncommon and potentially life threatening as it is often not clinically suspected. The ruptured spleen usually has an associated pathology with the etiological factors reported as being a) neoplastic disorders (30.3%); b) infectious disorders (27.3%); c) Inflammatory, non-infectious disorders (20%); d) drug and treatment related disorders (9.2%) and e) mechanical disorders (6.8%) in a systemic review of a traumatic splenic rupture by Renzulliet al [1]. In this review of 845 patients only 59 cases (7%) had a truly "normal spleen" [1] thus supporting the theory that the spleen that ruptures is often 'defective' and usually has an underlying pathology contributing to its rupture.

The neoplastic disorders that form the bulk of pathological lesions found in these ruptured spleens are primarily malignant and non-malignant hematological disorders, as the spleen is a native hematopoietic organ. Occasionally, primary neoplastic disorders such as angiosarcoma, peliosis, cysts or hemangiomas may be seen in upto 8.1% of cases [1]. Secondary metastatic lesions to the spleen are rare. There are many theories proposed as to why this is so including a) Anatomical/ mechanical theory based on features such as constant flow of blood, rhythmic splenic contractions, sharp take off angle of the splenic artery, lack of afferent lymphatic vessels and b) Micro environmental-including high concentration of angiogenesis inhibition factor associated with high density of splenic lymphoid cells with immune surveillance inhibitory factors: thus being "poor soil" for tumor deposits that prevent growth of micro metastatic foci within the spleen [2-5], Hence, splenic metastases from solid tumors defined asparenchymal lesions are considered exceptional.

The majority of splenic metastases occur in the context of disseminated multivisceral metastases with the common primary organs reported as breast, lung, colorectal, ovarian carcinomas and melanomas [2]. In a 25-year clinicopathologic study of 92 Chinese patients with secondary nonlymphoid splenic tumors, the most common primary tumor site was lung (21%) followed by stomach (16%), pancreas (12%), liver (9%) and colon (9%) [3]. Solitary splenic metastasis is a rare event with a reported incidence of 2.3-7.1% [6]. Isolated splenic metastases from lung cancer, a very rare occurrence, can be synchronous [4, 6-10] or metachronous [11-19] to the primary tumor. Most of these metastases are asymptomatic, being diagnosed on radiological imaging and often confirmed with fine needle aspiration or core biopsies as required [2]. Exceptionally, fine needle aspiration of a splenic mass can reveal a clinically occult primary tumor [20]. Infrequently, patients with splenic metastases can present with splenic rupture (12 %), abdominal pain (21%) and fever (3%) [6, 21-22].

Lung cancer is one of the most commonly diagnosed cancers and is the leading cause of cancer related death in Canada [23]. Based

on histology, lung cancer is divided into two main types: Small Cell Lung Cancer (SCLC) and the more prevalent (85-90%) Non-Small Cell Lung Cancer (NSCLC), which is comprised of adenocarcinoma (50%), squamous cell carcinoma (40%) and large cell carcinoma (10%) [6, 23].

Approximately 50% of all lung cancers are metastatic at diagnosis [5, 23]. The most common sites of metastases from NSCLC include the brain (47%), bone (36%), liver (22%), adrenals (22%), thoracic cavity (11%) and distant lymph nodes(10%) [23]. The spleen is an uncommon site of metastases with reported incidence in <5% and has been identified as an independent poor prognostic factor with poor outcomes [5-6].

Pathologically, splenic metastases can present as gross macroscopic or microscopic disease. In the microscopic pattern, there is no visible gross macroscopic lesion, and the tumor cells can be within venous sinuses, trabecular vessels and /or the red or white pulp. Splenic metastases can present as three main macroscopic/ gross patterns: i) macro nodular; here in, the splenic parenchyma is replaced by macroscopic identifiable solitary or multiple large nodules,

ii) micro nodular, characterized by scattered uniform military size nodules in the white or red pulp and iii)diffuse, wherein, the splenic parenchyma is replaced by tumor cells [2, 6, 11]. In our case the splenic rupture was associated with a macro nodular pattern of a single nodule. The pathological diagnosis of the mass being metastatic and of lung origin was confirmed by its typical histomorphological features of being an epithelial gland forming adenocarcinoma associated with the confirmatory pulmonary immunophenotype (CK7+, TTF1+, NapsinA+; CK20-and CDX2). Adenocarcinoma is the most commonly diagnosed histologic type of NSCLC and is also the most commonly diagnosed histologic type in people who have never smoked as seen in our case, who had no underlying risk factors for lung cancer. Recently, it has been proposed that Non–Small Cell Lung Cancer (NSCLC) is a different entity in people who never smoked than in people who used to smoke or currently smoke [23]. The splenic metastases reported in lung cancer are predominantly non-small cell cancers being composed of squamous cell carcinoma [10, 14-15, 17, 19] or adenocarcinoma [7-8, 12-13, 21], and a few poorly /undifferentiated /large cell carcinomas with splenic metastases [16, 18-19, 22] have also been reported.

Splenic rupture, a potentially life threatening event, is usually secondary to trauma. Non traumatic rupture of the spleen due to metastases is exceedingly rare [2-3]. In a 25 year clinicopathological study of 92 patients, 2% of patients had secondary non-lymphoid splenic metastases, with the most common primary lesion being metastatic choriocarcinoma and melanoma [3]. The exact cause of the mechanism of splenic rupture postulated in the context of malignancy include a) direct invasion of the splenic capsule by the malignant cells, b) pressure theory of necrosis within the neoplasm leading to intratumoral bleeding with resultant increased intrasplenic pressure leading to rupture and c) the prothrombotic nature of malignant cells leading to splenic infarctions -thus causing subcapsular bleeding and subsequent rupture. Though splenic metastases have been reported in lung carcinoma [4-19, 21-22, 24-25] and a few cases have been reported with rupture of the spleen during chemotherapy treatment [26-27], initial presentation of lung cancer with splenic rupture is an extremely rare event [1, 4, 24-25]. In these cases, the primary lung cancer was diagnosed prior to the splenic rupture [4, 25] and in one case the spleen was truly a 'spontaneous' rupture with no underlying pathological abnormality [24].

To the best of our knowledge, ruptured splenicmetastasis as the first presentation of a clinically occult disseminated primary lung cancer, as seen in our case has not been previously reported. Increased awareness with high index of suspicion of this extremely uncommon clinical presentation of an underlying common disease is important as rapid diagnosis can significantly reduce major morbidity and mortality.

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