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Doege-Potter Syndrome Secondary to a Solitary Fibrous Tumor of the Abdominopelvic Cavity: A Case Report

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Abstract **Case Report**

Solitary fibrous tumors (SFTs) are rare neoplasms initially identified as "localized fibrous mesothelioma" predominantly found in the pleural cavity. Common clinical manifestations include pain, the presence of a palpablemass, as well as neurological or vascular symptoms. Tumors located in the abdomen or pelvis can exert pressure on adjacent structures, leading to symptoms such as urinary obstruction or retention, bowel obstruction or constipation, and abdominal distention. Doege-Potter syndrome is characterized by hypoglycemia resulting from excessive production of insulin-like growth factors by the tumor. We report the case of a 54-year-old male patient, diagnosed with type II diabetes 8 years ago, currently not undergoing any treatment, Admitted for investigation of recurrent episodes of hypoglycemia.

Keywords: Solitary fibrous tumors, Doege-Potter syndrome, abdominopelvic mass, rare, imaging, diagnosis.

INTRODUCTION

Solitary fibrous tumors (SFTs) are rare neoplasms initially identified as "localized fibrous mesothelioma" predominantly found in the pleural cavity. Over time, the origins of SFTs have sparked controversy, leading to the current understanding of SFTs as ubiquitous fibroblastic-derived mesenchymal neoplasms [1]. These tumors are exceptionally rare, accounting for less than 2% of all soft-tissue tumors. The literature has reported only a limited number of cases involving extrathoracic localizations. In this study, we present a case involving a male adult patient with an abdominopelvic SFT. The objective is to elucidate the pathological and imaging characteristics associated with SFTs arising from the abdomen and pelvis [2].

CASE REPORT

A 54-year-old male patient, diagnosed with type II diabetes 8 years ago, currently not undergoing any treatment, Admitted for investigation of recurrent episodes of hypoglycemia. Physical examination revealed an abdominal mass laterally located on the left flank. Abdominal ultrasonography revealed a supravesical abdominopelvic mass that appeared hypoechoic, heterogeneous, with areas of necrosis, and showed vascularization on color Doppler.

To further investigate the patient's condition, an abdominal and pelvic CT scan was performed and identified a voluminous abdominopelvic tissular mass measuring 18 x 17 cm, with lobulated contours and relatively well-defined boundaries. The mass exhibits calcifications and areas of fatty density, and shows heterogeneous enhancement upon contrast administration

The mass displaced the bladder downward and appeared to infiltrate the seminal vesicles and the prostate. Anteriorly, it came into intimate contact with the left anterolateral abdominal wall. Posteriorly, it displaced the digestive structures, including the sigmoid colon, and came into contact with the vertebral bodies. No other associated abdominal or thoracic lesions were noted.

A germinal cell tumor or mesenchymal tumor were suggested as possible diagnosis based upon the imaging apearences. Histopathologic and immunohistochemical analyses after biopsy were consistent with a solitary fibrous tumor.

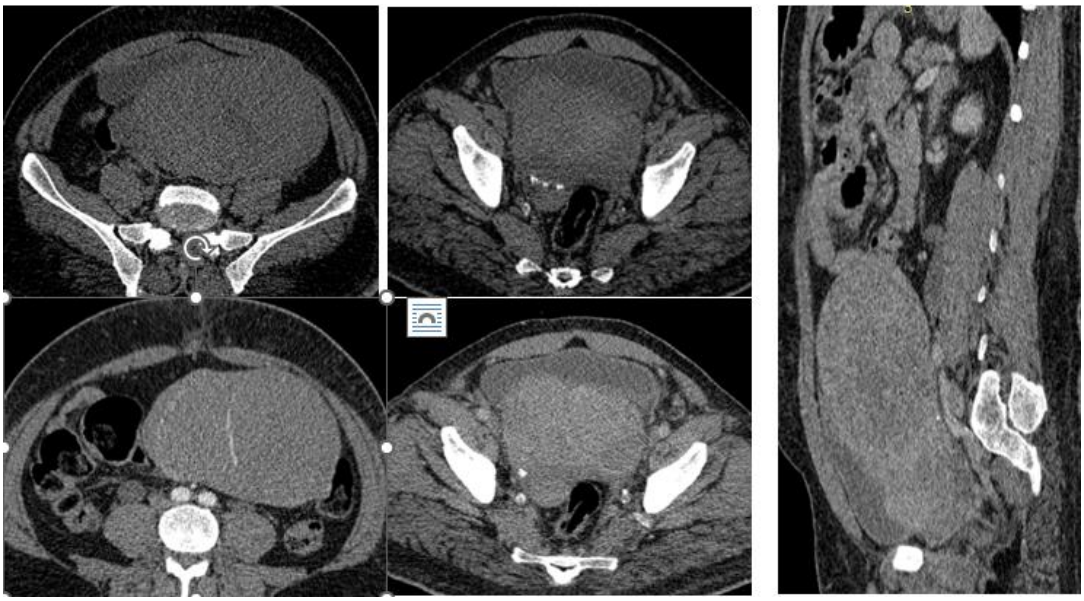

Figure: Abdomino pelvic CT scan showing a voluminous bdominopelvic tissular mass, with lobulated contours and well-defined boundaries. The mass contains calcifications and areas of fatty density, and shows heterogeneous enhancement upon contrast administration. The mass displaces the bladder downward and infiltrate the seminal vesicles and the prostate. Anteriorly, it came into intimate contact with the abdominal wall.

DISCUSSION

Solitary fibrous tumors (SFTs) are rare neoplasms of mesenchymal origin, constituting less than 2% of all soft-tissue tumors. Traditionally believed to originate from mesothelial cells and primarily affecting the pleura, pericardium, and peritoneum, recent advances in pathology have provided a deeper understanding of their histogenesis and remarkable tumor distribution heterogeneity. It is now recognized that SFTs are ubiquitous neoplasms originating from fibroblastic or myofibroblastic cells and can arise in various anatomical locations throughout the body [3].

Clinically, SFTs typically present as slow-growing masses in middle-aged adults and are often asymptomatic. There is an equal distribution of SFTs between men and women. Common clinical manifestations include pain, the presence of a palpable mass, as well as neurological or vascular symptoms. Tumors located in the abdomen or pelvis can exert pressure on adjacent structures, leading to symptoms such as urinary obstruction or retention, bowel obstruction or constipation, and abdominal distention [4].

A small subset of SFTs, accounting for less than 5% of cases, may exhibit a unique manifestation known as Doege-Potter syndrome, characterized by hypoglycemia resulting from excessive production of insulin-like growth factors by the tumor. This syndrome was observed in our case [5].

Extra pleural SFTs can occur in different areas of the body, including the head and neck (such as the meninges, orbit, upper respiratory tract, salivary glands, and thyroid gland), the abdomen and pelvis (including the liver, pancreas, peritoneum, retroperitoneum, gastrointestinal tract, mesentery, kidney, adrenal gland, urinary bladder, prostate, spermatic cord, and female genital tract), as well as other locations like the periosteum and somatic soft tissues [5]. Cross-sectional imaging modalities play a fundamental and essential role in the identification, characterization, and localization SFTs, while also provide valuable guidance for surgeons during operative procedures.

Cross-sectional imaging techniques play a pivotal role in the identification, characterization, and localization of solitary fibrous tumors (SFTs), while also providing crucial guidance for surgeons during operations. Computed tomography (CT) is the preferred initial diagnostic modality for detecting SFTs. In CT scans, SFTs typically manifest as well-defined, hypervascular masses that may induce displacement or exert pressure on adjacent organs, such as the bowel, urinary bladder, blood vessels, and ureter. The tumor may exhibit central regions of reduced contrast enhancement or no enhancement at all, which indicate areas of necrosis or cystic changes. Calcifications, although infrequent, can occur in larger benign or malignant tumors. CT imaging is also valuable for assessing the local extent of the tumor, including invasion into nearby structures, as well as identifying any locoregional or distant metastases [6].

Magnetic resonance imaging (MRI) is another imaging modality employed to evaluate SFTs. On T1-weighted MR images, SFTs typically exhibit intermediate signal intensity, while on T2-weighted images, they demonstrate heterogeneous low signal intensity with the presence of flow voids. Following the

administration of gadolinium contrast material, SFTs display marked enhancement [6]. The treatment of choice for SFTs is surgical excision. However, for unresectable SFTs, antiangiogenic therapy has shown promising preliminary outcomes. Neoadjuvant radiation therapy and systemic chemotherapy have also been attempted, albeit with varying success rates [7].

CONCLUSION

Recent advancements have contributed to a comprehensive understanding of the morphologic diversity of solitary fibrous tumors (SFTs). These neoplasms are now acknowledged as ubiquitous mesenchymal entities originating from fibroblastic precursors. While SFTs exhibit variable clinical behavior, as well as diverse imaging manifestations, a heightened awareness of the tumor distribution and imaging features can facilitate the diagnostic accuracy

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