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Squamous Cell Carcinoma Arising in a Mature Pelvic Teratoma in an Adult Male with Situs Inversus Totalis: A Rare Case Scenario

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ABSTRACT

Background: Extragonadal pelvic teratomas are extremely rare in adult males and may pose significant diagnostic and therapeutic challenges. Malignant transformation within a mature cystic teratoma is uncommon, with squamous cell carcinoma being the most frequent histological subtype. Association with situs inversus totalis is exceedingly rare.
Case Presentation: A 43-year-old man presented with a progressively enlarging abdominal lump for 7 years, with rapid increase in size over the preceding 2 months accompanied by abdominal pain, constipation, altered micturition, and significant weight loss. Clinical examination revealed a large firm abdominopelvic mass occupying all abdominal quadrants. Imaging demonstrated mature teratoma with malignant transformation. Incidentally, situs inversus totalis was identified. Tumor markers including AFP and β-hCG were within normal limits. The patient underwent exploratory laparotomy with adhesiolysis, excision of the cystic mass, and peritoneal lavage. Histopathology revealed mature cystic teratoma with squamous cell carcinoma transformation and metastatic deposits in the cyst base. Postoperative recovery was uneventful, and the patient was planned for adjuvant chemoradiotherapy.
Conclusion: Adult male pelvic teratomas are exceptionally rare and should be considered in the differential diagnosis of large pelvic masses. Rapid enlargement, large tumor size, and solid enhancing nodules may indicate malignant transformation. Complete surgical excision remains the cornerstone of management.

Keywords: Pelvic teratoma; Mature cystic teratoma; Squamous cell carcinoma; Extragonadal germ cell tumor; Situs inversus totalis

INTRODUCTION

Teratomas are germ cell tumors composed of tissues derived from more than one embryonic germ layer and arise from totipotent primordial germ cells. [1] Although commonly encountered in gonadal locations, extragonadal teratomas are uncommon and predominantly occur along the midline. Pelvic and retroperitoneal teratomas in adult males are exceedingly rare, accounting for less than 1% of all teratomas. [2]

Mature cystic teratomas are generally benign lesions; however, malignant transformation may occur in approximately 1–2% of cases, particularly in long-standing tumors. [3] Squamous cell carcinoma is the most frequently reported malignancy arising from mature teratomas. [4] Risk factors associated with malignant transformation include older age, large tumor size, rapid growth, and long duration of symptoms.

Situs inversus totalis is a rare congenital anomaly characterized by complete mirror-image transposition of the thoracic and abdominal viscera, resulting in their anatomical arrangement opposite to the normal configuration. [5] The coexistence of pelvic teratoma with malignant transformation and situs inversus totalis is extremely uncommon.

We report a rare case of squamous cell carcinoma arising in a mature pelvic teratoma in a 43-year-old male with situs inversus totalis, managed surgically with planned adjuvant chemoradiotherapy.

CASE SUMMARY

A 43-year-old man presented with a progressively enlarging abdominal lump for 7 years, with rapid increase in size over the preceding 2 months accompanied by abdominal pain, constipation, altered micturition, and significant weight loss. The patient was apparently well 7 years prior to presentation when he noticed a right lower abdominal swelling measuring approximately 5 × 5 cm. The swelling progressively enlarged and showed rapid increase in size over the preceding 2 months, reaching approximately 20 × 20 cm and causing abdominal distension and dragging abdominal pain. There was no history of fever, vomiting, melena, hematochezia, cough, shortness of breath, jaundice, or bony pain.

Clinical examination revealed a large firm abdominopelvic mass approximately 30 × 25 cm occupying all abdominal quadrants. The swelling was firm, smooth, non-tender, and had well-defined margins with limited mobility (figure 1). Percussion revealed dullness over the swelling. Bowel sounds were normal. Bilateral testes and spermatic cords were grossly normal.

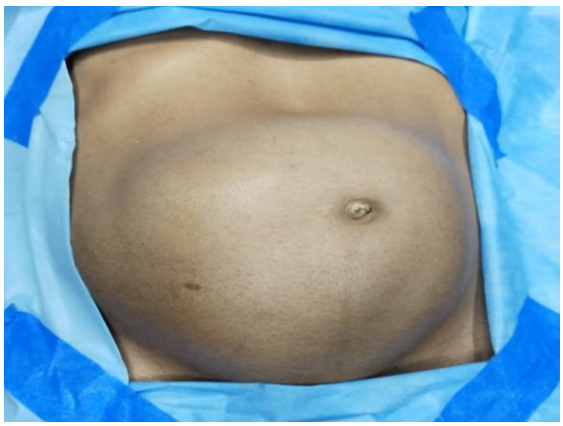

Fig 1 – Clinical examination

Imaging demonstrated a large heterogeneous cystic-solid abdominopelvic lesion with fat-fluid levels, calcifications, Rokitansky nodules, and multiple peritoneal deposits suggestive of mature teratoma with suspicion of malignant transformation. Incidentally, situs inversus totalis was identified (figures 2, 3). Tumor markers including AFP and β-hCG were within normal limits.

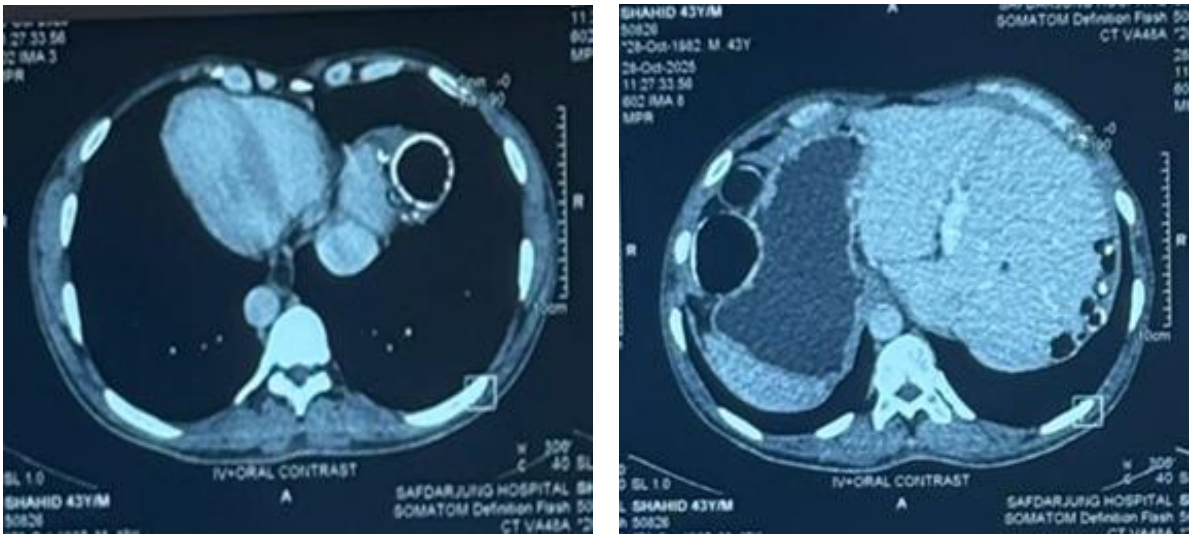

Fig 2,3 – Situs inversus

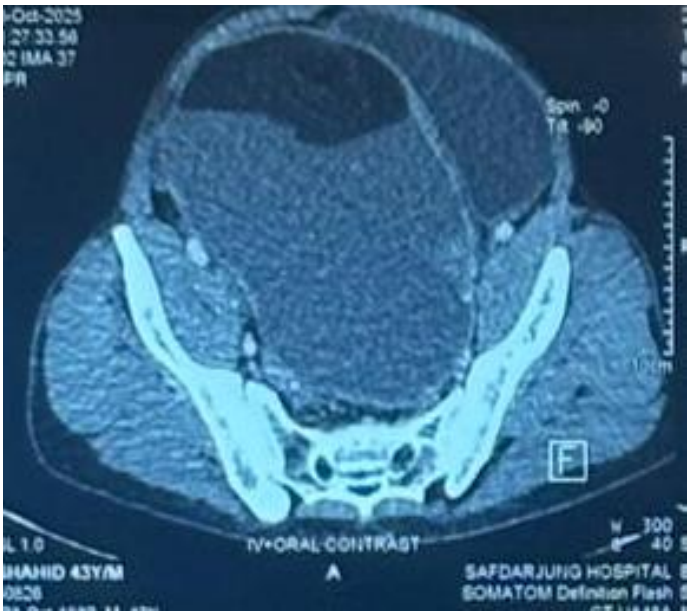

Fig 4 – Large abdominopelvic cyst

The patient underwent exploratory laparotomy with intraoperative findings suggestive of a large cystic mass measuring approximately 30 × 30 cm extending from the suprapubic region to the xiphisternum was identified (figure 4). Dense adhesions were present involving the liver, gall bladder, small bowel loops, urinary bladder, sigmoid colon, and rectum. Multiple cystic deposits were seen over the omentum and parietal peritoneum. Situs inversus anatomy was confirmed intraoperatively. Adhesiolysis, excision of the cystic mass, and peritoneal lavage was done.

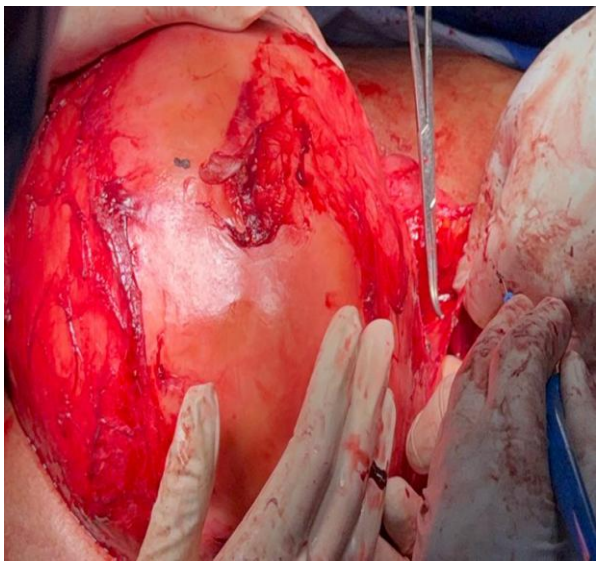

Fig 5 – Intraoperative cyst

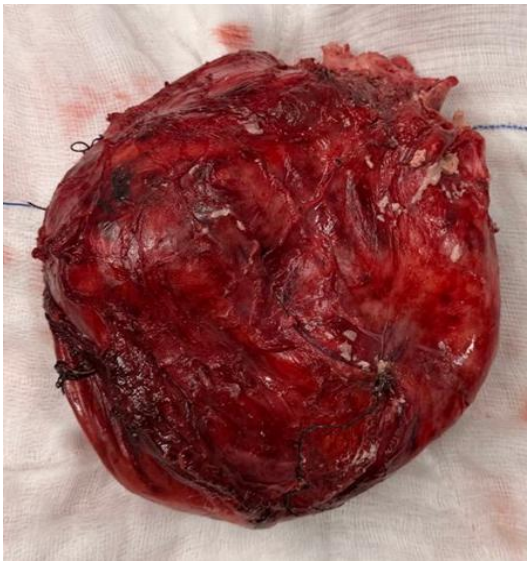

Fig 6 – Histopathology specimen

Histopathology revealed mature cystic teratoma with squamous cell carcinoma transformation and metastatic deposits in the cyst base (figures 5, 6). Postoperative recovery was uneventful, and the patient was planned for adjuvant chemoradiotherapy.

DISCUSSION

Extragonadal pelvic teratomas in adult males are exceptionally rare tumors. They arise from aberrantly migrated primordial germ cells that fail to reach the gonadal ridge during embryogenesis. These tumors are most commonly located in the sacrococcygeal region, mediastinum, retroperitoneum, and intracranial compartment.

Mature cystic teratomas are generally benign lesions; however, malignant transformation may occur in long-standing lesions. Squamous cell carcinoma represents the most common malignant transformation due to the predominance of squamous epithelium within mature teratomas.

Several radiological features may suggest malignant transformation, including large tumor size, rapid enlargement, mural nodules, solid enhancing components, and capsular irregularity. In the present case, the lesion demonstrated Rokitansky nodules, solid enhancing areas, and rapid interval enlargement, raising suspicion for malignancy.

Tumor markers such as AFP and β-hCG are usually normal in pure mature teratomas and help exclude mixed germ cell tumors. Testicular evaluation is recommended in male patients to exclude a primary gonadal tumor or a regressed ("burnt-out") testicular germ cell tumor. [8,9]

Complete surgical excision remains the mainstay of treatment for pelvic teratomas. [6,10] En bloc resection with avoidance of intraoperative spillage is recommended whenever feasible. Adjuvant therapy depends on the histological subtype of malignant transformation and should be individualized according to the malignant component. [7] Squamous cell carcinoma arising within teratoma often demonstrates poor response to conventional germ cell tumor chemotherapy, and platinum-based regimens may be considered in selected cases, although treatment should be guided by the histology and clinical context. [7]

The coexistence of situs inversus totalis further increased the operative complexity in the present case. Published pediatric teratoma data also support surgery and follow-up as central components of management, although those data are not directly generalizable to adult malignant transformation. [10] To the best of our knowledge, very few cases combining

pelvic teratoma, malignant transformation, and situs inversus totalis have been reported in the literature.

CONCLUSION

Pelvic mature cystic teratoma with squamous cell carcinoma transformation in an adult male is exceedingly rare. Rapid tumor enlargement, large size, and enhancing solid nodules should raise suspicion for malignant transformation. Cross-sectional imaging plays a crucial role in diagnosis and surgical planning. Complete surgical excision remains the cornerstone of treatment, while adjuvant therapy should be tailored according to the malignant histological component.

Declaration by Authors

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