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**Case Report** 

# Inguinoscrotal Swelling an Unusual Presentation of the Intra-abdominal Pathologies

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#### Abstract

Inguinal hernia and lymphadenopathy are the most common causes of the inguinal swelling which can be diagnosed clinically, but the intraabdominal pathologies presenting primarily as an inguinal swelling and masquerading as inguinal hernia are often rare. We report 3 cases of the different primary intraabdominal pathologies presenting atypically as inguinal swelling and causing a diagnostic dilemma. Thorough clinical history, examination and better imaging accurate preoperative diagnosis and plan of management. 1<sup>st</sup> case was gallstone related necrotizing pancreatitis presenting with left lumbar pain and left inguinal swelling due to collection tracked into inguinal canal through patent processus vaginalis, managed conservatively with percutaneous drain for collection. 2<sup>nd</sup> case was metastatic liposarcoma in the left inguinal region who initially underwent excision of retroperitoneal sarcoma and received adjuvant chemoradiotherapy, he underwent wide local excision. 3<sup>rd</sup> case was low grade appendiceal mucinous neoplasm presenting initially as right indirect inguinal hernia followed by left inguinal hernia due to the mucinous material tracking into inguinal canal through processus vaginalis. Managed by herniorrhaphy and right hemicolectomy.

**Keywords:** Inguinal hernia, Appendix mucocele, Liposarcoma, Pseudomyxoma peritonei

**Abbreviations:** LFT: Liver Function Test; AXR: Abdominal X-Ray; USG: Ultra Sonography; CECT: Contrast Enhanced Computed Tomography; PCD: Percutaneous Drain; FNAC: Fine Needle Aspiration and Cytology; EBRT: External Beam Radiotherapy; PET: Positron Emission Tomography; FDG: Fluoro-2-deoxy-d-glucose; CEA: Carcinoembryonic Antigen; MRI: Magnetic Resonance Imaging; PMP: Pseudomyxoma Peritonei

# Introduction

Acute pancreatitis is an acute inflammatory process of the pancreas. Mortality ranges from 3% in patients with interstitial edematous pancreatitis to 17% in patients who develop necrotizing pancreatitis. Presentation varies with severity like pain abdomen, local complication and/or organ failure. Management requires a multidisciplinary approach like medications, intervention and/or necrosectomy [1]. Mucinous tumor of the appendix grows and occludes the lumen, mucus accumulates and the appendix ruptures. The peritoneum is then seeded with mucus-producing cells that continue to proliferate and produce mucus. The progressive accumulation of the mucus results in abdomen distension which is the most common presentation in both sexes. 2<sup>nd</sup> most common presentation in males is inguinal hernia (25%) and in females with ovarian mass [2]. Presentation of necrotizing pancreatitis and appendix mucocele with inguinal swelling is rare, causing diagnostic dilemma hence detailed history, examination and better imaging make preoperative accurate diagnosis and plan of management. Retroperitoneal sarcoma often presents late due to large volume of intraabdominal space [3]. Rarely they have discontinuous lobulated growth patterns with inguinal extension, the cause for recurrence in inguinal region in our case might be due to non-excision of the small inguinal extension of the primary retroperitoneal sarcoma.

## **Case Presentation**

#### Case 1

A 60-year male patient presented with gallstone related mild pancreatitis, sonography revealed cholelithiasis with pancreatitis without

any collection, serum amylase, lipase, and LFTs were within normal limits managed conservatively. Asymptomatic for 4 weeks, presented with bilateral flank pain and left inguinal swelling. On examination fullness and tenderness in left flank region and  $3 \times 3$  cm firm tender solitary swelling in the left inguinal region, no cough impulse and appears to be obstructed inguinal hernia (Figure 1). AXR done, does not features suggestive of intestinal obstruction. USG showed cholelithiasis, left inguinal hernia with collapsed small bowel loops as contents, no evidence of dilated bowel loops. In view of discrepancy in clinical and sonography findings, CECT abdomen done which revealed heterogeneous pancreas with less than 30% necrosis, peri-pancreatic necrotic collection anterior to the body of the pancreas, extending bilaterally in to paracolic gutter, on left side the collection tracking in to the inguinal canal and appears to be organized (Figure 1). Diagnosis of gallstone related necrotizing pancreatitis with local complications without any organ failure was made. In view of symptomatic spreading necrotic collection, PCD was placed into bilateral paracolic gutter collection and draining the necrotic output and culture was sterile. Single time aspiration of the left inguinal collection yielded only 1cc pus due to the organized collection and culture was sterile. After 2 weeks of the pigtail drainage patient symptomatically improved and the left inguinal swelling size gradually reduced and disappeared. Right PCD removed in view of nil output and no residual collection. Left PCD draining thick necrotic brownish fluid, hence PCD upgraded as per step-up approach. The patient improved significantly with PCD management and was asymptomatic. After 3 months, presented with

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pneumonia and septic encephalopathy in emergency, CECT showed no evidence of significant abdominal collection and consolidation of bilateral lungs. Later succumbed to death due to sudden cardiac arrest in 3 days.

# Case 2

58-year male patient with known diabetic and hypertensive, presented with low-grade fever, pain abdomen in the left iliac fossa region, constipation and decreased frequency of micturition for 3 months. On examination, vague lump in left iliac region. Initial sonography was suggestive of retroperitoneal mass. CT imaging showed heterogeneously enhancing soft tissue mass of size  $11 \times 9$  cm in the left retroperitoneum region, anterior to psoas, displacing the sigmoid colon anteriorly, with peripheral increased vascularity and traction over the left spermatic cord. CT guided FNAC revealed poorly differentiated liposarcoma. On laparotomy revealed well-circumscribed 11 × 9 cm hard mass in the left iliac region extending into pelvis adhered densely to external iliac vessels but not infiltrating, related medially to sigmoid colon, laterally to psoas muscle, ureter was normal, external iliac lymphadenopathy noted largest 2  $\times$  1.5 cm. On cut section solid grey white homogenous mass without areas of hemorrhage and necrosis. Post-operative period was uneventful. Histopathology showed liposarcoma dedifferentiated type and margins were free of tumor. Completed adjuvant chemo-radiotherapy with 50 Gy of EBRT and 6 cycles of vincristine, doxorubicin and cyclophosphamide regimen. At 3 months CT imaging, no evidence of residual disease or recurrence. The patient lost to follow-up for period of one year. At 1.5 years after surgery presented with painless progressive swelling over left inguinal region for 3 months duration. On examination hard subcutaneous mobile swelling of size  $4 \times 5$  cm, no cough impulse. Clinically recurrence of the tumor with lymph nodal metastases was suspected. CECT abdomen showed no evidence of recurrence or residual disease at the previous surgery site, soft heterogeneous tissue in the left inguinal region encasing the spermatic cord suggestive of liposarcoma (due to recurrence at the inguinal site or new spermatic cord liposarcoma) (Figure 2). No evidence of lung or liver metastases noted. With high suspicious of recurrent tumor, FNAC done showed as metastatic liposarcoma. Later underwent PET-CT imaging which showed FDG avid (SUV max 21.1) left inguinoscrotal hernia with omental fat of size  $5.2 \times 5.0 \times 6.2$  cm (Figure 2). No other FDG avid lesion noted. Later patient underwent exploration and the findings were  $10 \times 10$  cm hard mass encasing the spermatic cord, no evidence of inguinal lymphadenopathy or intraperitoneal extension. Wide local excision of the tumor with spermatic cord and inguinal orchidectomy was done. Post-operative period was uneventful. Histopathology revealed the findings suggestive of metastatic liposarcoma dedifferentiated type.

# Case 3

65-year male patient presented with irreducible right inguinoscrotal swelling, on examination diagnosis of right-sided irreducible indirect inguinal hernia was made and underwent surgery. Intraoperatively the indirect sac noted filled with mucinous-gelatin material and sac contents evacuated and mesh hernioplasty done. Later CECT abdomen done which showed a multiloculated collection of size  $9.7 \times 4.7$  cm, with few septations, soft tissue components, and calcification was seen insinuating between bowel loops in right iliac fossa region. CEA was 10.5 ng/ml. Hence referred to our institution, during the course of follow-up developed left-sided reducible indirect inguinal hernia and was irreducible at later time. Planned for surgical intervention, on inguinal exploration  $6 \times 3$ cm indirect sac contained mucinous-gelatin material and sac was densely adhered to spermatic cord, spermatic cord was preserved, left herniorrhaphy done. Laparotomy exploration revealed thick viscid mucinous-gelatin material present in right iliac fossa and pelvis. Cauliflower shaped tumor arising from the tip of appendix was perforated (Figure 3). Omentum and other solid organs were normal. Surgical debulking and right hemicolectomy was done and post-operative period was uneventful (Figure 3). Histopathology revealed low-grade appendiceal mucinous neoplasm, margins, and retrieved lymph nodes were free of tumor. Adjuvant CAPEOX regimen 6 cycles given and at 6 months follow up CEA reduced to 1.95 ng/ml and CECT abdomen no evidence of tumor recurrence.



Figure 2: a): heterogeneously enhancing soft tissue mass (green arrow) in the left retroperitoneal region of size 11 × 9 cm displacing sigmoid colon towards right (red arrow); b): CT guided FNAC from the lesion, lesion shows loss of fat plane with psoas; c): Heterogeneous mass in the left inguinal region, spermatic cord is not separately identified compared to the right side; d): PET-CT shows fdg avid (SUVmax 21.1) lesion in the left inguinoscrotal region encasing the spermatic cord cord or oright side (right spermatic cord is free); e): Soft tissue tumor with predominant fatty tissue excised with wide margins (arrow denotes the medial side); f): Resected specimen shows cut section of soft tissue tumor (black arrow), and testis with cord structures (small arrow).s



Figure 3: a): Right hemi-colectomy specimen, cauliflower-shaped growth at the tip of appendix, base of appendix is free of tumor; b): Right side inguinal incision of 1<sup>st</sup> surgery, left side inguinal incision for evacuation of the indirect sac mucinous, midline incision for the right hemicolectomy.

#### Discussion

#### Acute necrotizing pancreatitis

Acute pancreatitis is an acute inflammatory condition, with a range of severity as well as various local and systemic complications. Gallstones and alcohol are the most common causes of acute pancreatitis [4]. A majority of patients have mild acute pancreatitis, in the subset of patients with organ failure (severe disease) or infected necrosis, the mortality rate reaches up to 30% [4]. Acute pancreatitis is divided into early and late phases. The early phase occurs in the 1<sup>st</sup> week after onset, with the disease manifesting as a systemic inflammatory response. The late phase, which generally starts in the 2<sup>nd</sup> week and can last for weeks to months, occurs only in patients with moderately severe or severe pancreatitis, as defined by persistent organ failure and by local complications such as pancreatic and peripancreatic collection [4]. collection may track along the fascial planes in the abdomen into various distant spaces and hence presentation may vary with the site of the collection.

Inguinoscrotal swelling occurring during acute pancreatitis is very rare, causing diagnostic dilemma [5,6]. The inflammatory fluid in the peripancreatic spaces may extend anteriorly into pararenal spaces and retro mesenteric plane, posteriorly into the retro renal plane, laterally into lateroconal plane, superiorly along the diaphragm to enter the mediastinum and inferiorly into the pelvic retroperitoneum. These fascial planes are weak barriers for enzyme-rich pancreatic fluid. Rarely, the fluid may track along the deep inguinal ring, inguinal canal, superficial inguinal ring in to the scrotum due to patent process vaginalis. And testis is spared from the inflammation due to the tunica vaginalis [7]. In our case collection was limited to the inguinal canal. In such cases it can mimic complicated inguinal hernia, testicular torsion, epididymo-orchitis, hydrocele, testicular tumor, etc [8,9]. a high degree of suspicious should be kept in such cases. CT imaging should be considered for accurate diagnosis and proper plan of management. The correct diagnosis is utmost important because the inguinal collection can be managed by non-surgical intervention and to avoid wrong surgical intervention [9]. Drainage of intra-abdominal collection

will cause resolution of the swelling or sometimes may require local drainage of collection and close follow up is required till the definitive treatment is given.

#### Liposarcoma

Liposarcoma is the most common type of soft tissue sarcoma that occurs most commonly in the extremities (59%), followed by torso (18%), retroperitoneum (13%), head and neck (9%) [10]. The majority of the liposarcoma that originates in the retroperitoneum occurs at 40-60 years of age with equal sex distribution (1:1) [3,10]. Retroperitoneal liposarcoma is asymptomatic at onset due to the large volume of intraabdominal space allows the tumor to grow without compressing the vital structures when symptoms present the tumor has usually grown very large with local invasion [3].

CT/MRI is required for diagnosis and differentiation of the retroperitoneal tumors. On CT scan, retroperitoneal liposarcoma usually appears as a large encapsulated mass containing variable amounts of fatty and soft tissue components [11]).

The final diagnosis of retroperitoneal liposarcoma is dependent on the pathological and immuno-histochemical analyses. Liposarcoma can be histologically subdivided into 5 subtypes: Well-differentiated, Myxoid, Round cell, Pleomorphic and Dedifferentiated [12,13]. Well-differentiated liposarcoma may recur locally, but the metastatic potential is low, while pleomorphic liposarcomas have high metastatic potential, which may reduce the survival rate [14]. Para-testicular tumors are benign (70%) and malignant (30%); the spermatic cord is the most common site of origin. The most common benign tumor is lipoma and common malignant tumors are sarcoma (as the spermatic cord is almost completely derived from the mesoderm) [15]. Liposarcoma and leiomyosarcoma are the most common sarcoma subtype in adults while MFH and fibrosarcoma are rare (about 10% and 5% respectively) [16].

The distinction of primary cord liposarcoma which arises in and confined to the inguinal canal and no connection with retroperitoneal fat, from the inguinoscrotal extension of a retroperitoneal liposarcoma is critical for management and determination of prognosis [14]. In our case, there was diagnostic dilemma whether the tumor was because of the recurrence of the primary tumor due to the discontinuous lobulated growth pattern with inguinoscrotal extension or the new tumor originating from the spermatic cord. The mainstay of treatment is radical surgical resection with a negative margin  $\geq 1$  cm. The margin status is one of the most critical variables that dictate further management and determines prognosis. Local recurrence remains the preliminary cause of mortality in retroperitoneal liposarcoma [17]. In our case the recurrence of the tumor in the inguinal region may be due to the retroperitoneal origin of the tumor with inguinoscrotal extension as a discontinuous lobulated growth (separate nodules), which was not evident in the CT scan done initially. This growth pattern of the tumor explains the recurrence of the tumor at the inguinal region with being tumor-free margin of the primary resected specimen as noted in our case [18].

#### Appendix mucocele

Neoplasms of the appendix are rare, found in approximately 1 percent of appendectomy specimens and account for only approximately 0.5%-1% percent of intestinal neoplasms [19,20]. Appendiceal mucocele refers to any lesion characterized by distended, mucus-filled appendix either benign or malignant. Appendiceal mucocele due to mucosal hyperplasia or retention cyst have indolent nature even if they rupture, they are not associated with recurrence. Appendiceal mucinous neoplasms are classified into mucinous adenoma (confined to mucosa), low-grade appendiceal mucinous neoplasm (glandular invasion confined to muscularis mucosae) and appendiceal adenocarcinoma (glandular invasion into muscle layer). Rupture of these appendiceal neoplasms leads to pseudomyxoma peritonei [2].

PMP is a unique condition characterized by the diffuse collection of gelatinous material in the abdomen and pelvis, peritoneum seeded with mucus-producing cells, which continue to proliferate and produce mucus, this progressive accumulation of copious amount of mucinous fluid gradually fills the peritoneal cavity, resulting in the characteristic jelly-belly [2]. the mucus tracks through the patent processus vaginalis into the inguinal canal and present as indirect inguinal hernia.

PMP is more common in females in 5<sup>th</sup>-6<sup>th</sup> decade, most commonly presents with abdominal distension, 2<sup>nd</sup> most common presentation in males is inguinal hernia as in our case and in females as ovarian mass [2]. The characteristic radiologic feature is a heterogeneous enhancing collection with scalloping of liver, spleen, mesentery and redistribution phenomenon [21].

There is still controversy in the treatment of the PMP. Periodical surgical debulking removes gross disease limits the buildup of mucus and its pressure effect. This treatment is not curative inevitable recurrence requires repeated surgical debulking [22]. Aggressive cytoreduction and intraperitoneal chemotherapy is an alternative to periodical debulking with a concept of the rarity of the extra-peritoneal spread of PMP and intraperitoneal high drug concentration compared to systemic administration might control the growth of peritoneal disease [23]. When mucinous material is encountered during hernia repair, recovery of the fluid and hernia sac for histologic examination is important [24].

Acute pancreatitis is an acute inflammatory condition, with a range of severity as well as various local and systemic complications. Gallstones and alcohol are the most common causes of acute pancreatitis [4]. A majority of patients have mild acute pancreatitis, in the subset of patients with organ failure (severe disease) or infected necrosis, the mortality rate reaches up to 30% [4]. Acute pancreatitis is divided into early and late phases. The early phase occurs in the 1<sup>st</sup> week after onset, with the disease manifesting as a systemic inflammatory response. The late phase, which generally starts in the 2<sup>nd</sup> week and can last for weeks to months, occurs only in patients with moderately severe or severe pancreatitis, as defined by persistent organ failure and by local complications such as pancreatic and peripancreatic collection [4]. collection may track along the fascial planes in the abdomen into various distant spaces and hence presentation may vary with the site of the collection.

## Conclusion

A detailed history, careful physical examination, keeping the possibilities of atypical causes and better imaging helps in accurate diagnosis and a proper plan of management of "An inguinoscrotal swelling with an unusual presentation of intraabdominal pathologies" to avoid unnecessary wrong surgical intervention.

#### **Conflict of Interest**

We declare that there is no conflict of interest regarding the publication of this paper.

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