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CASE REPORT

ASYMPTOMATIC EXTENSIVE PELVIC LIPOMATOSIS ASSOCIATED WITH SYMPTOMATIC EPIGASTRIC HERNIA: A RARE CASE REPORT

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ABSTRACT: INTRODUCTION: Pelvic lipomatosis is a rare benign disorder characterized by deposition of mature fat tissue in the pelvic cavity and absence of delimitation by a capsule. Peak incidence of this disease is between 25 to 60 years of age with a staggering male predominance of 10:1. **CASE REPORT:** A 45 yr. old male presented with epigastric swelling and abdominal pain. Clinically, he was diagnosed with epigastric hernia. Ultrasound showed bladder wall thickening with bilateral hydroureteronephrosis. CT showed bilateral hydrouretronephrosis with pear shaped bladder, non-encapsulated fatty mass surrounding urinary bladder symmetrically, with attenuation similar to that of subcutaneous fat suggesting extensive pelvic lipomatosis. On laparotomy there is a large pelvic lipoma filling the whole of pelvis and bladder was surrounded by fatty tissue all around along with lipoma of the cord. Patient underwent pre peritoneal hernioplasty for ventral hernia and excision of pelvic lipoma and the lipoma of the cord. **DISCUSSION:** Pelvic lipomatosis is a benign overgrowth of adipose tissue with small amount of inflammatory and fibrotic components found especially in the perivesical and perirectal spaces. The best definitive diagnostic procedure is CT. Various treatments have been attempted, but no single treatment has proved effective to date.
KEYWORDS: Pelvic lipomatosis, epigastric hernia, cystitis glandularis, hydroureteronephrosis.

INTRODUCTION: Pelvic lipomatosis is a rare beningn disorder characterized by deposition of mature fat tissue in the pelvic cavity and absence of delimitation by a capsule.[1] This condition was originally described by Engels in 1959[2] and was induced the term "pelvic lipomatosis" to define a clinical-radiologic entity by Fogg and Smyth in 1968.[3] Peak incidence of this disease is between 25 to 60 years of age with a staggering male predominance of 10:1.[1] It has been demonstrated that approximately three-quarters of cases with pelvic lipomatosis have histologic evidence of proliferative cystitis, such as cystitis cystica and cystitis glandularis, although its cause remains unknown.[4] It causes various symptoms due to compression of pelvic organs by intra pelvic overgrowth of mature fatty tissue.[5]

CASE REPORT: A 45 yrs. old male presented with complaints of an epigastric swelling since 30 years and abdominal pain since 8 years. Swelling was insidious in onset, gradually progressive. Pain started 8 years back, intermittent colicky in nature and aggravated by exertion. There are no urinary disturbances or any co morbidities. On examination, vitals are stable and there was a reducible epigastric hernia measuring 5x5 cms with bilateral direct inguinal hernias. All laboratory parameters were in normal limits except for urine analysis which showed mild pyuria. Urine culture grew no growth. Ultrasonography showed ventral hernia with fat as its content. 3x2cm ventral wall defect about 2 inches above the umbilicus.

Bladder wall thickening with bilateral hydroureteronephrosis, with pre-void residue 280 cc and no post-void residue. Computed tomography (CT), (Figure 1) showed bilateral

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hydrouretronephrosis with pear shaped bladder, non-encapsulated fatty mass surrounding urinary bladder symmetrically, with an attenuation similar to that of subcutaneous fat suggesting extensive pelvic lipomatosis. Patient underwent cystoscopy, which showed high bladder neck with inflamed bladder mucosa and left ureteric orifice edema. On exploratory laparotomy (Figure 2) there is a large pelvic lipoma filling the whole of pelvis and bladder was surrounded by fatty tissue all around along with lipoma of the cord. Patient underwent pre peritoneal hernioplasty for ventral hernia and excision of pelvic lipoma and the lipoma of the cord. Specimens are sent for histopathology (Figure 3) and diagnosis confirmed. Patient recovered well and is symptom free after 1 year of follow up.

DISCUSSION: Pelvic lipomatosis is a benign overgrowth of adipose tissue with small amount of inflammatory and fibrotic components found especially in the perivesical and perirectal spaces.[1] Etiology of the disease has not been established. However, it has been speculated that the fat proliferation might be associated with chronic pelvic inflammation due to chronic urinary tract infection.[6] Certain studies suggest that the etiology is possibly related to obesity but not related with diabetics.[1] But some researches indicated that obesity was not the single factor responsible for the pathogenesis and the narrow male bony pelvis may also contribute to its currency and development.[7]

Wilson et al[8] postulated that this entity could be secondary to a hormonal mechanism or hormonal metabolic alteration that induces the deposition of fat because it usually affects men in the fourth decade of life or older. Abbott and Skinnerys, [9] suggested that congenital venous anomalies and subsequent venous stasis were contributing factors. subsequent venous stasis was contributing factors. Histologically, the condition is characterized by diffuse overgrowth of mature fatty tissue, but this is often accompanied by cell infiltration and fibrosis suggestive of chronic inflammation.[5] Clinical features are due to varied compression phenomenon over the urinary tract (lower urinary tract symptoms), rectum (constipation, tenesmus, bleeding), vascular system (edema, lower extremities).[1]

Pelvic lipomatosis usually causes progressive obstructive uropathy by obstructing the ureters. This phenomenon may be an indication for radical cystoprostatectomy and urinary diversion.[10] edema. Diagnostically, no characteristic findings are present in the urine, and a palpable mass above the pubic bone, elevation of the prostate gland, and hypertension are frequent[5, 11], but not necessarily universal observations. Radiography is indispensable for the diagnosis of pelvic lipomatosis. In plain radiograms, intrapelvic fatty tissues are imaged as characteristic radiolucent areas.[10] By IVP, an inverted tear-drop shaped morphological change of the bladder with elevation of the base and elongation of the apex is a characteristic observation.[11]Evident hydronephrosis and hydroureter and medial deviation of ureters are occasionally observed.[11] The best definitive diagnostic procedure is CT.[6] CT images demonstrate deposition of fatty tissues causing compression of adjacent structures, possibly resulting in relevant morphological deformities.[6]

It is important to highlight the relevant role of CT urography in the demonstration of the typical presentation of pear-shaped bladder, which is generally accepted as a valuable characteristic indicative of pelvic lipomatosis[7], besides hydronephrosis due vesicoureteral obstruction. It is proposed that the optimal method to increase sensitivity to predict pelvic lipomatosis, especially in the patients with early stage, is based on a quantitative measurement of volume of pelvic fat. This requires a three dimensional imaging data and a precisely designed mathematical model.[12]

Exploratory laparotomy, which used to be performed for the diagnosis, yielded few characteristic findings in most cases and there can be as high as a 10% risk of post-operative thrombophlebitis owing to impaired pelvic venous return.[5] Some of the important differential

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diagnosis are lipoplastic lympadenopathy and idiopathic increased fat deposition and liposarcoma. In both, excretory urogram shows lateral displacement of the kidney and ureter.[5] Lymphangiopathy is the most useful method that distinguishes pelvic lipomatosis from lipoplastic lymphadenopathy. In lipoplastic lymphadenopathy, the lymphangiogram shows dilated, tortuous and bizarre pelvic lymph channels, and large foamy pelvic and para-aortic lymph nodes.[5] Cystoscopy is a must for all the cases. The pathological findings of bladder lesions in these patients are usually cystitis glandularis, cystitis cystic or cystitis follicularis.[13] The association of proliferative bladder disease and pelvic lipomatosis is based on speculation that obstruction in drainage o the bladder results in a proliferative cystitis because of an altered environment rich in protein fluid. Cystitis cystic or cystitis glandularis could potentially be premalignant lesions o adenocarcinoma of the bladder. An association between cystitis glandularis and adenocarcinoma of bladder has been reported. It is recommended that patients with cystitis glandularis and pelvic lipomatosis receive strict surveillance to detect any associated adenocarcinoma o the bladder.[13]

Various treatments have been attempted, but no single treatment has proved effective to date. Some cases of response to dietary therapies have been reported.[5] Most urologists use a conservative approach with the use of symptomatic therapy and follow up for cases with minor symptoms and no renal impairment. Certain modalities of treatment have been employed with little success and these include long term antibiotics, steroids, diet control and even radiotherapy. Occasionally, an upper tract urinary diversion is required for those with obstructive uropathy with worsening renal function and severe symptoms.[1] It has been well described that excision of the pelvic fat is difficult and time consuming but not impossible and response to surgery is good, but recurrence is possible.[14] Ultrasonic assisted lipectomy and re-implantation of ureters have been employed successfully in cases of obstructive pelvic lipomatosis.[15] Ya Peng et al[13] stated that surgical removal of adipose tissue around the bladder does not improve the symptoms or radiological findings.

CONCLUSION: We report a rare case of pelvic lipomatosis with bilateral hydroureteronephrosis and symptomatic epigastric hernia. CT is the best diagnostic modality and in asymptomatic patients conservative approach is the most preferred approach.

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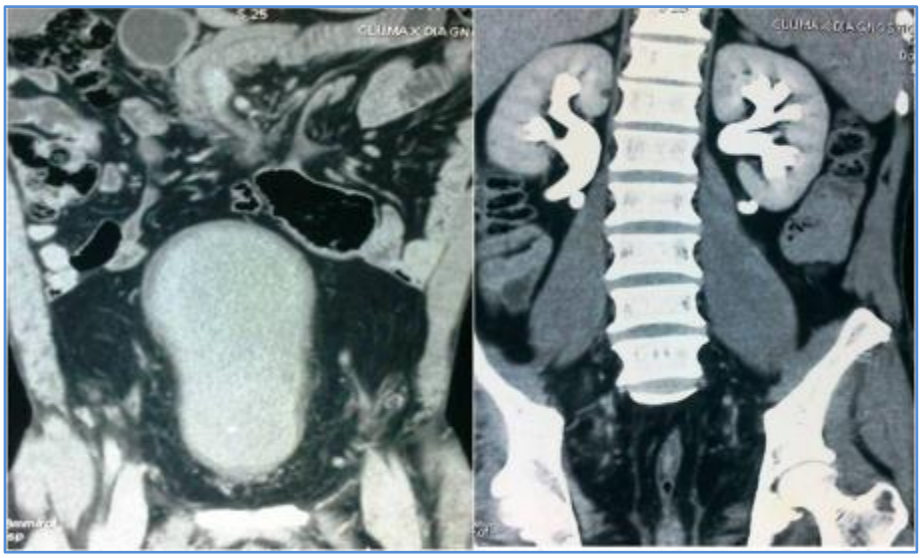

Fig. 1: CT Scan A. Pear Shaped Bladder B. Bilateral Hydroureteronephrosis

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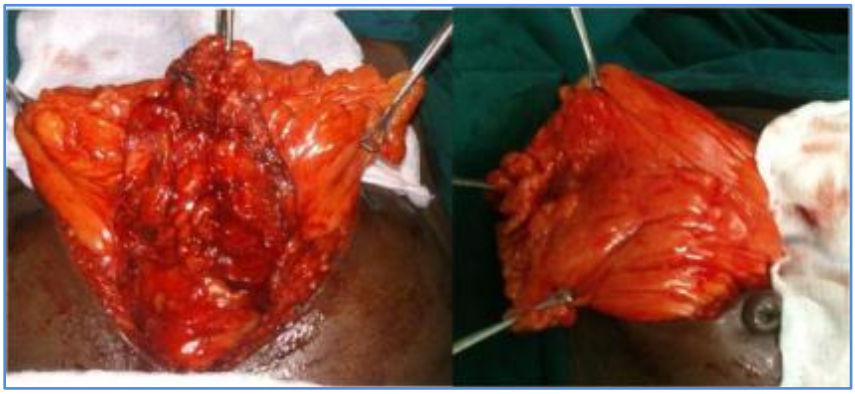

Fig. 2: Lipoma surrounding the bladder

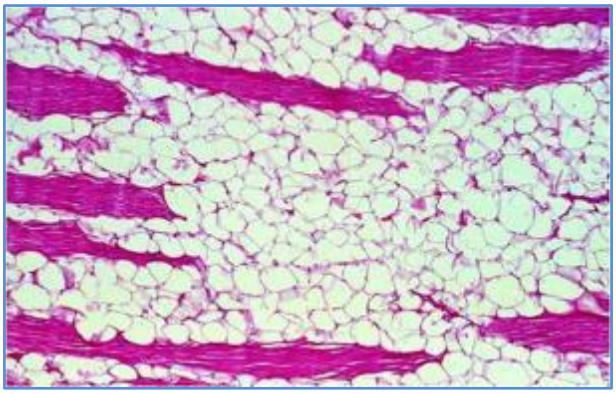

Fig. 3: Histopathology showing lipomatous elements

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