Original Research Article

Management of rare and atypical inguinal hernias: our experience at a tertiary care centre

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ABSTRACT

Background: Inguinal hernia is one of the most common surgical conditions operated by the surgeon. The purpose of the study paper was to provide diagnostic and therapeutic resources to deal with certain difficult situations in hernia repair.

Methods: A retrospective analysis of 8 rare and atypical inguinal hernias was conducted for a period of 2 years at NRI General Hospital, which includes cases like Loss of domain hernias and hernias with atypical contents; which stood as diagnostic and management challenge to the surgeon.

Results: Of the eight very rare and atypical cases operated, no postoperative complications were noted except one which developed a scrotal abscess after one month of discharge.

Conclusions: The infrequent encounter with rare and atypical hernias stands a diagnostic challenge to the surgeon. Inguinal hernia repair even-though looks simple yet may sometimes be very difficult.

Keywords: Inguinal hernia, Loss of domain hernia, Mesenteric cyst, Ogilvie's hernia, Ureteric hernia

INTRODUCTION

Hernia meaning rupture in Latin, is defined as an abnormal protrusion of an organ or tissue through a defect. It can occur at various sites in the body, most commonly the abdominal region.¹ Abdominal wall hernias occur only at sites where the aponeurosi and fascia are not covered by striated muscle. Inguinal hernia is the most common abdominal wall hernia.

Hernias are a common problem a surgeon comes across in his life. Approximately 75% of abdominal wall hernias occur in the groin.² Of these, inguinal hernia repair ranks among one of the most commonly performed surgical procedures. More than 20 million groin hernia repairs are performed every year worldwide.³ Inguinal hernias present along a spectrum of scenarios. Around 30% of these cases present asymptotically, and upto 50% of

them are unaware of the hernia.³ Few cases present with emergent symptoms not related to the hernia. As hernia is an elective procedure unless strangulated or obstructed, it does not catch the eye of a surgeon in those situations.^{1,2}

Herein we present certain rare and atypical presentations of inguinal hernia, which stood as a diagnostic and therapeutic challenge to us. However, the gold-standard repair of inguinal hernia is to perform a tension-free hernioplasty, which prevents further future recurrence in these cases.

METHODS

This was a retrospective analysis was conducted at NRI Medical College and General Hospital and of the various presentations of inguinal hernias operated at our institute, 8 cases were found to be rare and atypical in their

presentation. The data of the 8 cases were collected from our medical records, and followed retrospectively for a period of 24 months from August 2017 to August 2019.

Inclusion criteria

Eight rare and atypical inguinal hernia presentations which stood as diagnostic and surgical challenge were included.

Exclusion criteria

Normal presentations of inguinal hernias which had no diagnostic and surgical difficulties were excluded. Strangulated and incarcerated hernias were excluded.

The history and physical examination findings were noted. Radiological investigations were performed for these cases as per requirement to confirm the diagnosis and their management was planned accordingly.

RESULTS

Of the 8 cases presented, most (87.5%) of them were within the age range of 40-60 years. All the 8 cases presented atypically and had swelling in the groin region. Ultrasonography of groin region was done for diagnosing the groin swelling in 7 cases which was done only for confirmation.

The presentation of the 8 different types of hernias was rare in terms of their infrequent occurrence and atypical in terms of their defect and contents of the sac. Of 8 cases, 2 (25%) were atypical in terms of their defect that is one had a giant defect and one case had defect in transversalis fascia. And two (25%) cases had atypical contents, one with ureter and one with a mesenteric cyst. The remaining 4 cases (50%) were rare presentations of the normalcy. Their clinical characteristics are presented in the Table 1.

Table 1: Presentation of various rare and atypical hernias at our institute.

Type of hernia	Age/sex	Clinical presentation	Diagnostic modality	Management
Loss of domain hernia	44/M	Right inguinoscrotal swelling extending upto mid-thigh	Clinical and USG	Tension free hernioplasty
Ogilvie's hernia	45/F	Swelling in the right iliac region above the inguinal region	Clinical and USG	Tension free hernioplasty
Hernia with ureter as content	60/M	Inguinoscrotal swelling since 1 year with increased frequency of micturition since 1mth Past history of bilateral inguinal herniorrhaphy done 10 years ago	USG abdomen and scrotum; but CECT abdomen and scrotum was confirmatory	Tension free hernioplasty
Hernia with mesenteric cyst as content	23/M	Swelling in right side of groin since 3 months associated with history of trauma lump in the lower abdomen on the right side since 1 month	USG and MRI	Exploration, excision of cyst and inguinal herniorrhaphy
Hydrocele en bisac presenting as subacute intestinal obstruction	55/M	Subacute intestinal obstruction with bilateral transilluminant scrotal swellings	USG abdomen and Scrotum; CECT abdomen	Exploratory laparotomy
Hydrocele of hernial sac with bilateral inguinal lymphadenopathy	50/M	Swelling in the left Groin region since 1 year.	USG Inguinoscrotal and abdomen	Left tension free hernioplasty with left inguinal lymph node excision biopsy
Bilateral inguinal hernia with bilateral undescended testes with umbilical hernia	42/ M	Swelling in the umbilical region since 2 years associated with a swelling in the right inguinal region since 1 year	USG abdomen; CECT abdomen in view of extra peritoneal mass superior to bladder on USG	Right tension free hernioplasty; right orchidopexy with umbilical hernioplasty left orchidopexy planned after 6 months)
Bilateral inguinal hernia with femoral hernia in female	52/ F	Bilateral inguinal region swelling since 2 years known diabetic since 4 years	Clinical	Bilateral tension free hernioplasty with repair of right femoral hernia through inguinal approach

M-male; F-female; B/I- bilateral; Mth- month; H/o- History of; USG- ultrasound; CECT- contrast enhanced Computed tomography; MRI – magnetic resonance imaging.

Table 2: Postoperative outcome in present study.

Type of hernia	Total no. of cases	Post-operative complications					Postoperative duration of stay (in days)
		Seroma	Postoperative myalgia	Wound dehiscence	Mesh infection	Hernia recurrence	
Loss of domain hernia	1	1	1	1	0	0	10
Ogilvie's hernia	1	0	0	0	0	0	5
Hernia with Mesenteric cyst	1	1	0	0	0	0	7
Ureteric hernia	1	0	0	0	0	0	5
Hydrocele en bisac	1	1 (cord edema)	0	0	Not placed	0	10
Hydrocele of hernial sac	1	1	0	0	0	0	5
Bilateral inguinal hernia with undescended testis	1	1	0	0	0	0 (under follow up)	7
Femoral hernia	1	0	0	0	0	0	5
Total (%)	8	6 (75%)	1 (12.5%)	1 (12.5%)	0	0	Average- 6.7

All cases healed well postoperatively and were discharged uneventfully. Out of the 8 cases, six cases (75%) had wound seroma, which were managed by regular dressings during their hospital stay. After discharge, all cases were followed at every 15-day interval for the first 3 months and then monthly for the next 9 months. One case (12.5%) with the loss of domain hernia presented with a scrotal abscess one month after discharge, which was treated with incision and drainage. The reason could be due to the huge defect and secondary to the history of diabetes. There was no recurrence noted in 7 cases for the one year follow-up period, and one case is still under follow-up since 6 months, which has not reported any recurrence. 50% of the patients were discharged on 5th postoperative day, while 25% required a stay upto 10th postoperative day. Thus, the postoperative outcome of the cases was as described above as Table 2.

DISCUSSION

Hernia is an abnormal protrusion of part or whole of abdominal contents through an abnormal opening in the wall of the cavity which contains it.¹ Surgical treatment is complete when the defect is closed efficiently, and weak wall is strengthened. The content of hernia varies depending on the site of hernia. The literature describes many rare and atypical presentations of inguinal hernias. These cases were rare in that they occurred infrequently and atypical in terms of the defect or the content. A few of these presentations have been experienced at our institute which challenge the surgeon skills clinically and surgically.

Loss of domain hernia

Giant inguinal hernias are usually the result of neglect

and fear of surgical procedure. They are common among the rural populations. The defect is usually large (>10 cm), and the contents vary from small and large bowel, bladder, ovaries and rarely even stomach. Even though the defect is large, the occurrence of small bowel obstruction and strangulation are also reported in the literature.⁴ The chronicity of the disease poses a challenge both intraoperatively while reducing the hernial contents back to abdomen, and postoperatively due to increase in the intra-abdominal pressure causing a reduction in venous return, and decrease in tidal volume. Recurrence is also significantly higher; hence tension free hernioplasty is a must.

Figure 1: Loss of domain hernia. (A) Giant bilateral inguinal hernia extending upto mid-thigh with buried penis, (B) large hernia sac (c) small and large bowel seen as contents of the sac.

We experienced similar presentation of a patient, who had a complaint of swelling in the right inguinoscrotal region since 5 years (Figure 1). In view of chronicity of the hernia, the patient was pre-operatively assessed for respiratory and cardiac performance status. He was started on preoperative nebulization and incentive spirometry to increase the tidal volume as he was a

known smoker for about 25 years. Intraoperatively, almost the large and small bowel contents were noted within the hernia sac, and there was no necrosis of the bowel. We thus recommend that presence of such large swelling and huge content should not panic a surgeon. A prior preoperative preparation and postoperative care would help the surgeon in the battle of reducing the contents.

Ogilvie's hernia

It is a rare presentation of the tubular or the funicular type of direct inguinal hernias and is also known as prevesical hernia as the bladder may rarely present as content. The usual defect in direct hernia is within the transversalis fascia whereas in Ogilvie's hernia it is within the conjoint tendon.^{6,7} In this type, as the neck is small, the risk of strangulation is very high.

We report a case of a female who presented with a swelling in the inguinal region (Figure 2) which could not be appreciated on inspection due to the abdominal fat, but a cough impulse and a defect was noted on palpation just above the right inguinal region. Intraoperatively, the defect was within the conjoint tendon which, was repaired and mesh was placed within the inguinal region. Review of literature reports few such cases of Ogilvie's hernia dated as old as 1939 by Gill et al.⁵ Similar presentation was described as Busoga hernia by Elena et al as it was common among the people of Busoga area from Uganda.⁸

Figure 2: Ogilvie's hernia. (A) Swelling in the right inguinal region, (B) & (C) defect in the conjoint tendon.

Hernia with atypical contents

The usual contents within a hernial sac are omentum and intestine. Others being the bladder, Meckel's diverticulum, appendix and fallopian tube, and rarely ovary in females.⁹ These may appear as separate contents or may slide along the wall of the hernia sac as a sliding hernia.

But occurrence of ureter as a content was first reported in 1880, and from then <140 cases have been reported in the literature, with even fewer described with incarceration and ureteric obstruction.¹⁰ This is usually common among

the renal transplant cases.⁵ It is more common on the right side than the left side. Ureteral inguinal hernias are predominantly indirect rather than direct (80% versus 20%).⁹ Two variants have been described in literature-a) Para peritoneal (80%)- which is acquired and occurs as a sliding type; they have peritoneal covering and are mostly likely to be obstructed. b) Extra peritoneal (20%) are usually congenital and do not have a peritoneal covering.¹¹

The literature also reports few cases wherein the intra-abdominal malignancies and cystic lesions appearing as contents of the hernia, due to the weakness in the abdominal musculature.¹² These are the usual presentations of ventral hernias. But, there are situations where they may descend to the scrotum as one of our case containing a mesenteric cyst.

Figure 3: Right inguinal hernia with ureter as content. (A) ureter noted along the wall of the hernia, (B) & (C) depicting ureter with the cord structures.

In two of our cases, there were atypical contents noted within the hernial sac. One of these was a recurrent hernia with a previous history of bilateral inguinal herniorrhaphy 10 years ago, with complaints of increased frequency of micturition for 1 year. He had swelling in the right inguinal region since 1 year, which on examination was confirmed to be an inguinal hernia. On ultrasound, the ureter was identified as a content which was confirmed on a CECT abdomen. Intraoperatively, the right ureter was identified sliding along the wall of the hernial sac (Figure 3).

Another case presented for a complaint of abdominal mass in the right lower abdomen since 1 month (Figure 4). He gives a history of swelling in the right groin region since 3 months, which followed a history of trauma. On examination, the swellings in the groin and inguinal region were in continuity and were non-transilluminant and mobility of the swelling in the abdomen was restricted. Ultrasound abdomen and scrotum were suggestive of hematoma. MRI abdomen was planned to confirm the diagnosis, and it revealed a large cystic lesion in the abdomen herniating through the inguinal region into the right scrotum. Exploratory laparotomy with a midline incision along with inguinal herniorrhaphy through an inguinal incision on the right was planned.

Intraoperatively, a mesenteric cyst arising 20 cm proximal to the ileocecal junction extending into right scrotum was identified with adhesions between the small bowel and cyst wall, and there was altered fluid in the cyst.

Figure 4: Hernia with mesenteric cyst as content. (A) clinical image showing swelling in the right lower quadrant of abdomen and another swelling in the right scrotum, (B) MRI abdomen showing hypoechoic cystic lesion in the right lower part of abdomen extending into the right scrotum, (C) intraoperative image showing abdominal and scrotal components of the swelling, (D) Intestines adherent to the cyst could not be separated, (E) altered blood within the cyst.

Mesenteric cysts are rare with incidence of 1/1,05,000-2,50,000 in hospitalized adult surgical patients. Most of the mesenteric cysts remain asymptomatic, and up to 40% are diagnosed incidentally during surgery.¹³ About one third of the mesenteric cysts present as acute abdominal condition like intestinal obstruction, volvulus, and the rupture of the cyst. Mesenteric cyst presenting as an irreducible hernia is a very unusual case, and only 7 cases have been reported in the literature.^{14,15} These cases not only pose a challenge during surgery but also challenge the clinical acuity of the surgeon.

Rare presentations of hydrocele

Hydrocele en Bisac: It is described as hydrocele extending into the abdominal cavity forming two intercommunicating compartments (inguinoscrotal and abdominal).¹⁶ It is a rare variety seen the pediatric age group.¹⁵

We report a 55-year male with hydrocele en bisac and the presence of a closed loop obstruction makes it even more rare presentation. CECT abdomen revealed a closed loop obstruction of 4cm segment of the bowel around which a

collection of fluid was identified tracking down into the scrotum and presenting as hydrocele which was suggestive of abdomino-scrotal hydrocele/hydrocele en bisac. Exploratory laparotomy was planned and intraoperatively bilocular hydrocele noted with the sac in the abdomen containing 4 cm segment of the mid-ileal loop with adhesions to the sac causing a closed loop obstruction (Figure 5). The abdominal sac contained serous fluid, and this was in connection with the scrotal component. Dupuytren first described abdominal scrotal hydrocele and termed it as hydrocele en bisac in 1834.¹⁴ Since then, very few cases of hydrocele en bisac were described and this condition accounts for only 0.17% cases of different types of hydrocele. To the best of our knowledge, there were no cases of hydrocele en bisac presenting as closed loop obstruction.

Figure 5: Hydrocele En Bisac presenting as subacute intestinal obstruction. (A) Abdominal component seen as multiple loculated cystic swelling, (B) loop of ileum entering the cystic swelling in the abdomen, (C) adhesions between the sac and the ileal loop, (D) 4 cm segment of ileum within the abdominal part of the hydrocele en bisac, (E) abdominal part in continuity with the scrotal swelling as indicated by the forceps.

Figure 6: Hydrocele of hernial sac. (A) Clinical image with swelling in the left scrotum and the left groin with a small cystic swelling projecting out near the left testis, (B) serous fluid being drained from the cystic lesion surrounding the left testis which was in continuity with the hernia sac, (C) indirect hernial sac.

Hydrocele of hernia sac: It is defined as hydrocele associated with hernial sac. In present case, the presence of cystic nodule over the testis and inguinal lymphadenopathy mislead the diagnosis, while intraoperatively, hydrocele was noted along the indirect hernia sac.

Hernia with undescended testis

Undescended testis is usually noticed in the children and is diagnosed within the first few years of life, but is occasionally not noticed until adolescence or even adult life. Bilateral presentation is usually associated with sterility in adult life. The risk of testicular malignancy increases as age increases in case of undescended testis if left untreated.¹³ An associated indirect hernia or interstitial hernia is frequent, and is as high as 80%. In present case, despite the congenital absence of gonad within the scrotum, neither the patient nor his family seemed to be particularly concerned with the fact, which led to the delayed presentation in an older age. As the length of the cord was adequate and as the testis was normal in dimensions, orchidopexy was considered on the right side with a second stage orchidopexy for the left side after 6 months. There were very few cases, of an adult cryptorchid male with an inguinal hernia, reported to the best of our knowledge.

Figure 7: Bilateral inguinal hernia with bilateral undescended testis. (A) Scrotal hypoplasia with swelling in the right inguinal region and umbilical region, (B) hernial sac with omentum as content and cord structures could not be identified, (c) testis noted at the deep inguinal ring after reducing the omentum into peritoneum, (d) cord structures noted at the deep ring.

Hernia in females

The incidence of inguinal hernia in the females is 2% with a male to female ratio of 20:1. The incidence of femoral hernia in the females is common than in the males.^{1,2} However, the occurrence of bilateral inguinal hernia with femoral hernia in females is an uncommon presentation. We report a case of 52-year old female with

the complaint of bilateral inguinal region swellings since 2 years associated with a cough impulse. On examination, there were three swellings noted in the bilateral inguinal regions and the right femoral region, all associated with cough impulse. Intraoperatively, bilateral tension-free hernioplasty was done with the ligation of femoral sac.

Figure 8: Bilateral inguinal hernia with femoral hernia in female. (A) Swelling in the bilateral inguinal regions and right femoral region, (B) hernial sac in the right inguinal and femoral region.

CONCLUSION

The infrequent encounter with rare and atypical hernias stands a diagnostic challenge to the surgeon. The surgeon must always be alert regarding the possibility of atypical and rare contents during the surgical exploration of inguinal hernia. Inguinal hernia repairs, even though look simple yet, may sometimes be very difficult. Hence, a surgeon should be aware of anatomy of inguinal region which helps in handling a situation with ease.

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