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Research Article

Management of various rare and atypical hernias: experience at a tertiary care centre in central India

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ABSTRACT

Background: Abdominal wall hernias are most frequently encountered in surgical practice accounting for 15% - 18% of all surgical procedures. The incidence of abdominal wall hernia in different countries varies from 100 -300/100000 per year. Of which most common being the inguinal hernias and femoral hernia being the least common. Midline ventral hernias are the next common variety of abdominal wall hernia after inguinal hernia. Ours is a tertiary care hospital, medical college and PG institute in central India. Apart from routine hernias, we have managed successfully some very rare varieties and developed management protocols. Here we are providing an analysis of these unusual presentations.
Methods: This is a retrospective study in which we analyzed the data's of hernia patients presented in our institute over the period of 7 years (2008-2014). Out of 765 patients, who were diagnosed having hernia, 680 were operated in our institute. Among these cases, we have selected 12 very rare and unusual cases which stood as diagnostic and management challenge and which were operated by a single surgeon and his team.
Results: Out of the 12 cases of atypical hernias, 11 patients underwent surgical intervention. There was no mortality. 6 patients had some postoperative complications which were managed during their Hospital stay only. After discharge, all the patients were followed at regular intervals. There was no recurrence or other complication noted in 1 year follow up. Most common post-op complication was seroma which was seen in 50% of the operated patients. Other complications were wound infection (16.66%) and wound dehiscence (8.33%).
Conclusions: The infrequent encounter with the unusual varieties of Hernias can lead to the mental bias and becomes a diagnostic challenge for the general surgeon. The purpose of publication of these rare varieties is to increase our spectrum of knowledge and to provide diagnostic and therapeutic armamentarium to deal with these difficult situations.

Keywords: Hernia, Supra pubic cystotomy, Enterocutaneous fistula, Femoral, Amayand's

INTRODUCTION

Hernia is derived from a Latin word meaning "a rupture". Abdominal wall hernias are most frequently encountered in surgical practice accounting for 15% - 18% of all surgical procedures.[1,2] Worldwide, more than 20 million hernias are operated per year.

The incidence of abdominal wall hernia in different countries varies from 100-300/100000 per year.[3] Of which most common being the inguinal hernias and femoral hernia being the least common. Midline ventral hernias are the next common variety of abdominal wall hernia after inguinal hernia, most common after interventions involving the lower abdomen (incisional hernia). According to their locations, these are further

classified into umbilical, paraumblical and epigastric hernia. Traumatic and obturator hernias are very rare.[4]

Thoracic hernia is a very rare disorder which is mainly described in the context of trauma or following thoracic surgery.[5,6] Only a few cases are reported in literature till date.

Ours is a tertiary care hospital, medical college and PG institute in central India. Apart from routine hernias, we have managed successfully some very rare varieties and developed management protocols. Here we are providing an analysis of these unusual presentations.

The main objective is to enlighten the upcoming surgeons about the management of various rare and atypical hernias in order to improve their outcomes.

METHODS

This is a retrospective study in which we analyzed the data's of hernia patients presented in our institute over the period of 7 years (2008-2014). Out of 765 patients, who were diagnosed having hernia, 680 were operated in our institute. Among these cases, we have selected 12 very rare and unusual cases which stood as diagnostic and management challenge and which were operated by a single surgeon and his team. These were Suprapubic cystostomy (SPC) site hernia, Inguinal hernia presenting as spontaneous Faecal Fistula, Recurrent Incisional Hernia presenting as multiple enterocutaneous fistulas, Femoral hernia, Amayand's hernia, Intercostal hernia. All the patients were investigated with USG or CT scan as per requirement to confirm the diagnosis and their management was planned.

RESULTS

Out of the 12 cases of atypical hernias, 11 patients underwent surgical intervention. There was no mortality. 6 patients had some postoperative complications, which were completely managed during their Hospital stay only. After discharge, all the patients were followed at every 15 days interval for the first 3 months, and then monthly for the next 9 months. There was no recurrence or other complication noted in 1 year follow up.

Table 1: Showing comparison of various atypical hernias and their management.

Type of hernia		Age (years)	Sex	History of previous surgery	Diagnostic modality	Management
SPC site hernia	Case-1	65	M	SPC 3 years back (for obstructive uropathy)	USG-2.8cm defect at the SPC site	Hernioplasty with polypropelene mesh with creation of neo SPC site
	Case -2	45	M	SPC 5YRS back (for urethral injury	USG -2.2cm defect at the SPC site	
Femoral hernia	Case-1	40	F	None	Diagnosed intraoperatively	Lotheissen repair
	Case-2	38	F		USG	
	Case-3	55	M		USG	
Inguinal hernia presenting as fecal fistula	Case-1	23	M	None	USG & CT	1) Laparotomy with Resection of involved intestine. 2)Open drainage of inguinal area
	Case-2	35	M	I & D	USG& CT	
Incisional hernia with enterocutaneous fistula	Case-1	38	M	1) Laparotomy 10 years back 2)Incisional hernia surgery 6 years back	CT Sinogram	Laparotomy with Resection of involved intestine& Anatomical repair (fascial component separation) of Hernial site.
	Case-2	50	F	1) 2 Cesarean sections.(5 & 3 years back) 2) Incisional hernia surgery(1 year back)	CT	
Amayand's hernia	Case-1	55	M	None	Intraoperatively	Appendicectomy and Hernioplasty
	Case-2	61	M			
	Case-3	45	M			
Thoracic hernia	Case-1	60	M	None	HRCT scan Chest	Conservative (as the patient was unfit for any intervention)

The following cases and their management have been discussed in detail.

In both the cases of SPC site Hernia, previous SPC tract and hernia sac were isolated and excised, a new SPC tract was created 5 cm below and lateral to old site, hernia defect was closed, anterior rectus sheath was reconstructed and wide synthetic polypropylene mesh was placed.

Inonecase of Femoral hernia, the clinical and radiological diagnosis was inguinal hernia and the correct diagnosis of femoral hernia was established intraoperatively only. In the remaining 2 cases, femoral hernia was diagnosed through clinical and radiological methods. Lotheissen repair was done in all cases.

Two cases of inguinal hernia with fecal fistula were dealt in our hospital. Out of which one presented after Incision & Drainage of neglected obstructed inguinal hernia which was misdiagnosed as inguinal abscess and the other one presented with spontaneous fecal fistula. These two cases were managed in a similar manner with exploratory laparotomy and resection of obstructed fistulous ileal loopswith end to end Ileo-ileal anastamosis. Herniorrhaphy was done and inguinal area left opened for healing by secondary intention.

Two cases of incisional hernia with enterocutaneous fistulas were treated. One had the history of previous laparatomy and incisional hernia surgery, and the other had the history of previous 2 Caesarean sections and incisional hernia surgery. Exploratory laparotomy with resection of involved part of small intestine and end to end anastamosis of normal bowel was done. In first case, the previously placed mesh was found eroding into the bowel lumen and it was removed. Anatomical reconstruction of anterior abdominal wall was done in both the cases.

Three cases of Amayand's Hernia were managed. Intraoperatively, there was caecum with inflamed appendix as the content of hernia sac in both the cases. Appendicectomy with Hernioplasty was done.

One case of very rare Thoracic hernia was diagnosed. A60 year old male presented with a bulge over left chest wall since one year. The swelling was moving with respiration and coughing. He didn't have any history of thoracic surgery or chesttrauma. USG& HRCT chest showed a protrusion of lung through the intercostal space during valsalva and coughing. He was a known case of chronic obstructive pulmonary disease with severe pulmonary hypertension. Intervention was not done because patient was not fit for surgery.

Table 2: Showing post-op complications, postoperative stayand herniarecurrences.

Type of Hernia	Total no of cases	Post-op complications				Average post-operative stay(days)	Recurrences after 1 year
		Seroma	Infection	Wound dehiscence	Mesh infection		
SPC site hernia	2	1	0	0	0	8	No
Femoral hernia	3	2	0	0	-	5	No
Inguinal hernia presenting as fecal fistula	2	1	1	0	-	10	No
Incisional hernia with enterocutaneous fistula	2	1	1	1	-	15	No
Amayand's hernia	3	1	0	0	0	5	No
Total	12	6	2	1	0		-

Above table highlights the post operative complications and overall hospital stay after surgery. We found that the most common complication was Seroma formation that was seen in 6 out of 12 patients operated (50%). Wound infection was seen in 2 patients (16.66%), while wound dehiscence was seen in only 1 patient (8.33%).

The polypropylenemesh was placed in 5 of the 12 operated patients but mesh infection was not seen in any of them.

Out of two patients of inguinal hernia with fecal fistula, one patient developed Seroma while the other had wound infection. Out of the two patients of incisional hernia with enterocutaneous fistula, one patient developed only Seroma, and the other developed wound infection and later on, wound dehiscence also.

In cases of femoral and amayand's hernia , the only complication seen was Seroma formation that developed in 2 out of 3 femoral hernia cases and 1 out of 3 Amayand's hernia cases.

DISCUSSION

A hernia is the bulging of part of the contents of the abdominal cavity through a weakness in the abdominal wall. Surgical treatment is complete when the defect is closed efficiently and weak wall is strengthened. A content of a hernia varies depending on the site with the viscera that is present within the wall. Review of literature shows many unusual hernia presentations.

SPC site hernia

Incisional hernia from suprapubic catheter site is a rare but important complication. Possible causal factor for hernia may be the trocar traversing peritoneum before entering in to the distended bladder. Once the distended bladder returned to pelvis, it provides an opportunity for the peritoneal contents to herniate through the potential weakness of the trocar site.[7] Harlingam et al[8] reported a case of strangulated incisional hernia after SPC. But in our cases, both were uncomplicated hernias with sac lying very close to fibrosed SPC tract and adhered to it. Incisional hernia usually occurs after major abdominal surgery like laparotomy or abdominal hysterectomy. Hernia occurring after SPC insertion is very rare. We recommend that the new SPC tract should be made away from the incision site to prevent further recurrence after SPC site Hernia surgery.

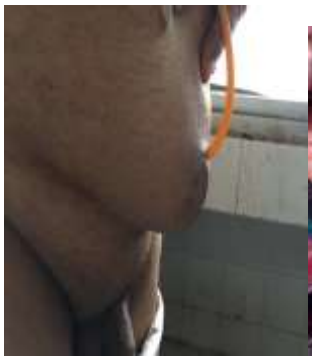

Figure 1 (a): Pre op.

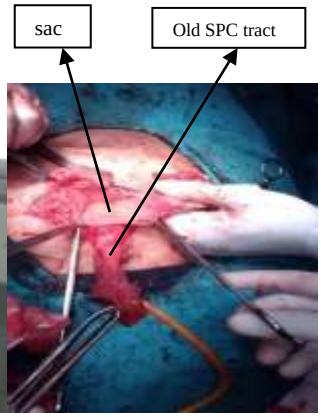

Figure 1 (b): Intra op.

Figure 1 (c): Intra op.

Figure 1 (d): Post op.

Figure 1: SPC site hernia.

Femoral hernia

A femoral hernia is an uncommon, acquired condition with incidence <5% worldwide, most commonly on right side. Berliner et al[9] Recommended repair of a reducible femoral hernia by using inguinal approach and of an irreducible femoral hernia by infra inguinal approach. If presence of incarceration or strangulation dictates need for emergent operation, the pre-peritoneal approach is preferred method. In one of our cases, we found the unusual inguinal presentation of femoral hernia on left side which was a serious diagnostic dilemma and final diagnosis was made on the operation table only. Our case highlights the importance of maintaining the patience and temperament when an unsuspected diagnosis is encountered during surgery.

Figure 2 (a): Pre op.

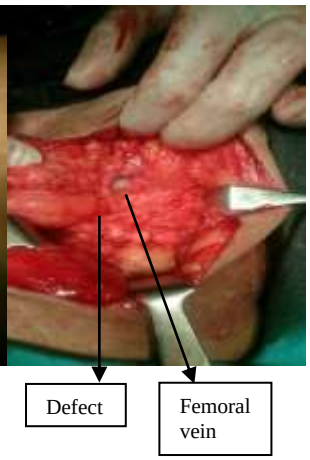

Figure 2 (b): Intra op.

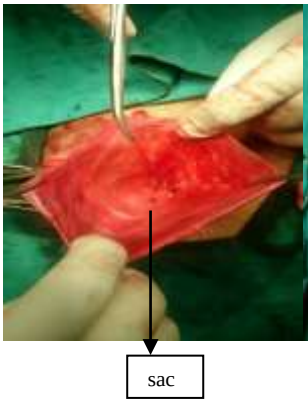

Figure 2 (c): Intra op.

Figure 2 (d): Immediate post op.

Figure 2: Femoral hernia.

Inguinal hernia with fecal fistula

Inguinal hernia presenting with faecal fistula mostly results from Richter's hernia. In 1598, Fabricius Hildanus[10] reported the earliest known case of a Richter's hernia. Richter's hernia is named after the German surgeon, August Gottlieb Richter, who gave the first description of this type of hernia in 1778. Onakpoya et

al[11] from Nigeria reported the case of a neglected Richter's inguinal hernia presenting with perforation and Fournier's gangrene. Three cases of spontaneous perforation of Richter's inguinal hernia with Fournier's gangrene were reported by Guzzo et al[12] in 2007 from the United States of America. One of our patient presented with spontaneous onset fecal fistula without any previous signs and symptoms of hernia and the other was a neglected undiagnosed obstructed inguinal hernia which was misdiagnosed as an inguinal abscess at the periphery and disastrous I & D was done. These finding were very unusual and final diagnosis was established by CECT abdomen and surgery was done. We do not recommend the use of mesh in the setting of abscess or fecal fistula. A good tightening of deep ring during laparotomy and free drainage from fistula site leads to healing and recurrence is prevented.

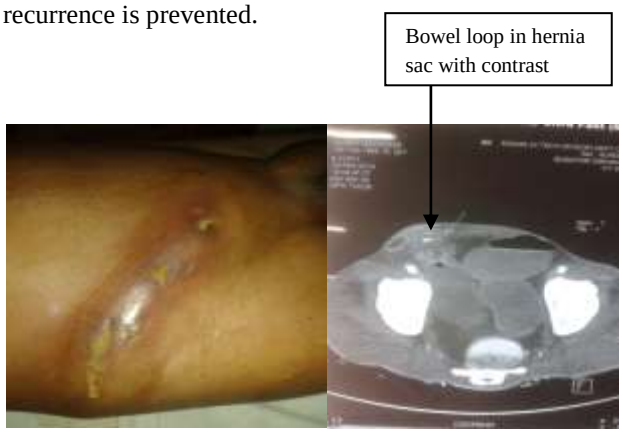

Figure 3 (a): Pre op. **Figure 3 (b): CT scan.**

Figure 3 (c): CT scan. **Figure 3 (d): Post op.**

Figure 3: Inguinal hernia as fecal fistula.

Incisional hernia with enterocutaneous fistula

Mesh hernia repair is a fairly common technique used to treat incisional hernias. Meshes have advantages and disadvantages. Whether meshes can cause enterocutaneous fistula, is a matter of debate. There are some studies which are against this possibility,[13,14] but there are also some studies in support of it.[15,16] In our case series, both the patients presented with recurrence of hernia along with enterocutaneous fistula few years after mesh repair for incisional hernia. In the first case, the incisional hernia had developed after Laparotomy, and in the other, the incisional hernia had developed after previous 2 Caesarean Sections. This suggests that a mesh can erode the bowel and that patients have a lifelong risk of this complication. We recommend the removal of whole involved mesh along with the resection of the involved part of intestine in such setting. The Anatomical repair of hernia can be done in such cases where the use of mesh is not indicated.

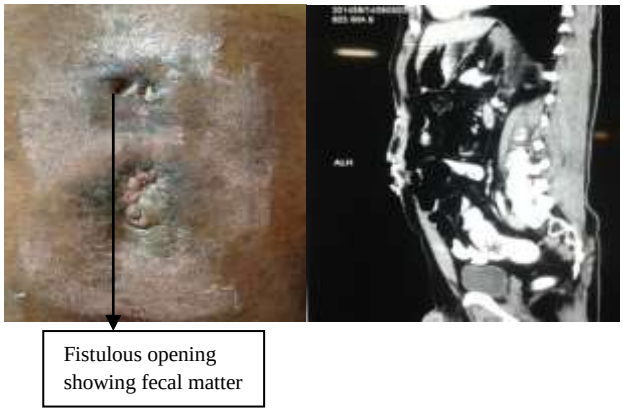

Figure 4 (a): Pre op. **Figure 4 (b): CECT.**

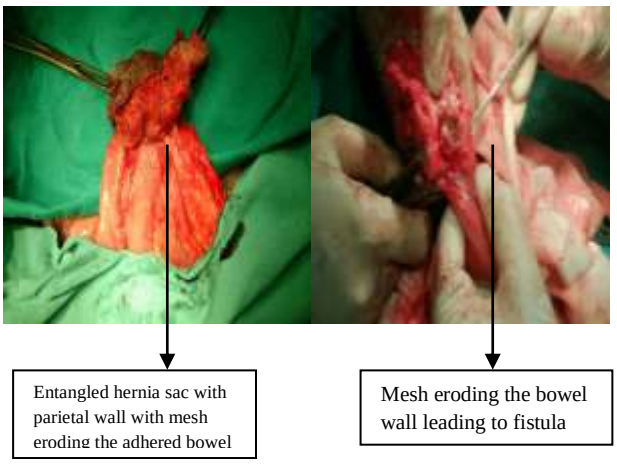

Figure 4 (c): Intra op-1. **Figure 4 (d): Intra op-2.**

Figure 4: Recurrent incisional hernia presenting as multiple enterocutaneous fistula.

Amayand's hernia

Amayand's hernia is named after Claudius Amayand (1680-1740), a French refugee and English surgeon who did the first recorded successful appendectomy in a case of hernia with appendicitis. When a hernial sac contains vermiform appendix, inflamed or un-inflamed, it is called as an Amayand's Hernia. True incidence of this condition is difficult to estimate as all the cases are not registered and reported. Ryan WJ et al[17] have reported only 11 cases of appendicitis out of 8692 (0.13%) cases of hernial sacs with appendix. D. Alia et al[18] observed presence of

appendicitis in 0.8% of 1341 operations of inguinal hernia. Vermiform appendix can be safely removed and hernia can be repaired in the usual way. In our cases Appendix and caecum was content of hernia sac and appendix was acutely inflamed. It's a matter of controversy that whether to put mesh or not when inflamed appendix is found. We have done hernioplasty because though appendix was inflamed, but there were no signs of perforation or pus formation.

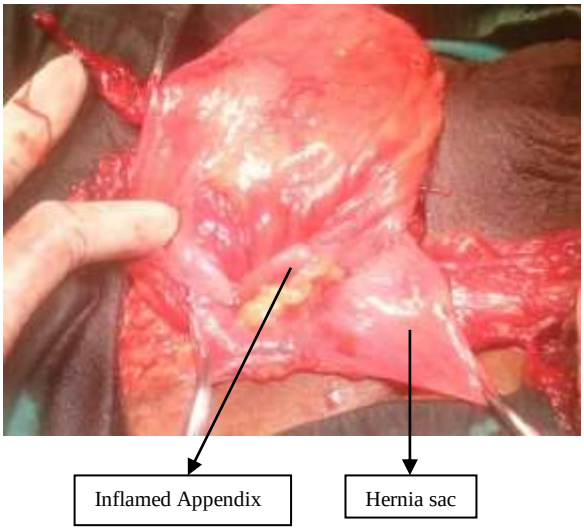

Figure 5: Intra-op pic showing appendix in hernia sac.

Thoracic hernia

Figure 6 (a) **Figure 6 (b)**

Figure 6: Thoracic hernia.

Thoracic hernia is a distinctly rare event, regardless of its location and cause. Most patients present hernia of the lung, defined as a protrusion of lung tissue beyond the confines of the chest cavity.[5,6] In contrast, the protrusion of soft tissue into the pleural cavity (inverted intercostal hernia) is extremely rare. In a study by Seder et al [19].showed twenty-seven consecutive patients underwent chest wall herniorrhaphy. Hernias most commonly occurred on the right side, in the fifth intercostal space, contained lung, and were chronic in nature. Pain was the presenting symptom in all except 4 patients. The most frequently observed co morbidities were obesity, chronic obstructive pulmonary disease (COPD), oral steroid use, and diabetes mellitus. Prosthetic herniorrhaphy is not associated with an increased risk of postoperative complications relative to primary repair. The treatment of underlying chest condition is of paramount importance and the successful outcome of hernia repair primarily depends on it.

CONCLUSION

The general surgeons, in their practice, frequently deal with common Hernias. Sometimes it leads to the mental bias and the infrequent encounter with the unusual varieties becomes a diagnostic challenge. The purpose of publication of these rare varieties is to increase our spectrum of knowledge and to provide diagnostic and therapeutic armamentarium to deal with these difficult situations.

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