SUBMITTED 13 OCT 20
REVISION REQ. 9 DEC 20; REVISION RECD. 11 JAN 21
ACCEPTED 10 FEB 21
ONLINE-FIRST: APRIL 2021
DOI: https://doi.org/10.18295/squmj.4.2021.068

Abdominoscrotal Hematocele in an Adult and Its Successful Treatment

***Uday S. Kumbhar, Oseen Shaikh, Sandeep Bhattarai**

Department of Surgery, Jawaharlal Institute of Postgraduate Medical Education and Research, Puducherry, India
**Corresponding Author's e-mail: k26uday74@yahoo.co.in*

Abstract

Abdominoscrotal hydrocele (ASH) is a variant of hydrocele rare to occur in adults. ASH has two sacs, one in the scrotum and one in the abdomen connected through the inguinal canal. Abdominoscrotal hematocele is a rare complication of ASH. We present a 57-year-old male patient, presented to us in August 2019 to JIPMER, Puducherry, India, with complaints of swelling in the scrotum for 15years and abdominal pain for two months. Both the swellings were soft, and cross fluctuation was present. Imaging confirmed the diagnosis of ASH. We did diagnostic laparoscopy, and the abdominal sac was decompressed and found to have thick brownish fluid suggestive of hematocele. We could demonstrate that both sacs were connected. Due to difficulty in the dissection of the sac, the procedure was converted to an open procedure. Both the sacs were excised, and Lytle's repair was done for the dilated internal ring.

Keywords: Testicular Hydrocele, Scrotal Hydrocele, Hydrocele, Hematocele, Testicular Hematocele, Scrotal Hematoceles, Laparoscopic Surgical Procedure, Laparoscopic Surgery

Introduction

Abdominoscrotal hydrocele (ASH) is defined as hydrocele extending into the abdominal cavity-forming two intercommunicating compartments.[1] ASH, also called hydrocele en-bisac, is a rare variant of a hydrocele that can occur commonly in the pediatric age group but rarely seen in the adult population. Patients are usually asymptomatic, or they can present with

variable sized abdominoscrotal swelling. ASH can get complicated due to bleeding inside the sac, which can occur due to trauma, malignancy, or spontaneous leading to the formation of hematocele.[2]Exact etiology of the ASH is not known, but there are various theories proposed. These theories explain disease occurrence either as an extension of scrotal sac into the abdomen or abdominal sac into scrotum or both. It is diagnosed clinically, and imaging is used to support the diagnosis.[3] Treatment can be conservative or surgical. In adults, there is no role of conservative management, and treatment is by surgical options, either open or laparoscopic approach.

Case Report

A 57-year-old male patient presented in August 2019 to JIPMER, Puducherry, India, with left-sided scrotal swelling for 15 years and abdominal pain for two months. There were no other symptoms like altered bowel or bladder habits. The patient was a non-alcoholic, non-smoker and didn't have any comorbidities. On examination, there was a large left side scrotal hydrocele and large left lower abdominal swelling (Figure 1). Abdominal swelling was soft, non-tender, and was having a smooth surface. Scrotal swelling was fluctuant; there was a cross fluctuation between abdominal and scrotal swelling. The transillumination test was negative in the scrotal swelling.

Ultrasonography of the abdomen and scrotum showed a large hypoechoic collection with internal echoes of size >20cm in the left lower abdomen with extension into the left inguinoscrotal region with normal bilateral testis. Contrast-enhanced computed tomography (CECT) abdomen showed a large hypodense retroperitoneal cystic lesion measuring 23 x 15 cm occupying the entire left side of the abdomen with the craniocaudal extent from the left lobe of the liver up to the pelvic brim. Anteriorly it was extending up to the anterior abdominal wall displacing entire intraperitoneal structures to the right side. Another hypodense cystic lesion was present in the left hemiscrotum measuring 17 x 9 cm with few septations and communication with abdominal cyst diagnostic of ASH or hydrocele en-bisac (Figure 2). The bilateral testis was appearing normal however size of the left testis was a little smaller than the right.

Diagnostic laparoscopy was done. Pneumoperitoneum was created by open technique with 10 mm trocar in the right lumbar region. Two working ports were used; one 5 mm trocar placed in the right lumbar region 7cm above and lateral to the umbilicus and another 10 mm trocar 6

cm below the umbilicus in the midline. There was a large retroperitoneal cystic lesion occupying almost the left half of the abdomen. It was displacing the sigmoid and descending colon medially. Cranio-caudally it was extending from the left lobe of the liver up to the pelvic brim. Inferiorly the cyst was extending into the inguinal canal through the deep inguinal ring. Due to the large size of the cyst and inadequate access for the dissection, a decision was taken to decompress the cyst. Initially, the cyst was punctured using a Veress needle passed transcutaneously. It was not possible to do effective aspiration as the fluid was thick and brown to greenish. Veress needle was replaced with 5mm trocar, and 1500 ml fluid was aspirated. Another 10 mm trocar was placed through the bottom of the scrotum into the hydrocele sac (Figure 3A), and 650 ml of the fluid was drained, which was of the same character as the abdominal fluid. There were two openings at the cranial pole of the sac, one of which was communicating with the abdominal cyst (Figure 3B). The retroperitoneal cyst was dissected from the lateral abdominal wall. Once the dissection proceeded medially, it was impossible to separate the cyst from the sigmoid colon and its mesentery. Because of the risk of inadvertent injury, the decision was taken for open conversion. The retroperitoneal cyst was approached transabdominally using a left inguinal incision. The cyst was opened, and brownish muddy fluid was evacuated. The cyst wall was completely excised. It was extending into the scrotum through the inguinal canal. A separate vertical incision was made over the scrotum. The hydrocele sac was opened, and the content was similar to that of the abdomen suggestive of hematocele. Testis also had a hematoma. A decision was taken for the left orchidectomy. Both the abdominal and scrotal sacs were dissected, and communication between the two sacs was well demonstrated (Figure 3C). Excision of both the sacs along with intercommunicating tract was done completely. The internal ring was mildly dilated; however, there was no weakening of the abdominal wall. Hence, we did the Lytle's repair for the dilated internal ring. Port sites, inguinal and scrotal incisions were closed.

The patient was discharged after eight days without any complication. The patient was under regular follow up. Operative site wounds healed completely, and the scrotum regained its normal size (Figure 4). We could find no recurrence of any swelling in the abdomen or scrotum at the end of one year follow up. We obtained written informed consent from the patient to use the images for publication purposes.

Discussion

Hydrocele means an accumulation of fluid between the layers of tunica vaginalis. It can occur in infants or adults. An Infantile hydrocele is due to persistent patent processus vaginalis. Adult or vaginal hydrocele occurs when fluid collection occurs between tunica vaginalis, where processus vaginal is obliterated. One more variant of hydrocele called hydrocele of cord occurs due to partial closure of processus vaginalis. ASH is a rare variant of hydrocele characterized by scrotal hydrocele extending into the abdomen as cystic abdominal swelling through the inguinal canal.

ASH is defined as hydrocele extending into the abdominal cavity-forming two intercommunicating compartments.[1]Dupuytren first described it in 1834, and it was called Dupuytren hydrocele or Hydrocele en-bisac.[1]Bickle in 1919 proposed a new term for hydrocele en-bisac called Abdominoscrotal Hydrocele.[2]

ASH is a rare entity and has been reported both in infants and old age. It is seen more commonly in the pediatric age group than adults. Overall in adults, there are less than 250 cases reported in the literature. India has the highest incidence of ASH in the world.[3] Literature review suggests that ASH is more common on the right side than the left. ASH is usually a congenital anomaly. It starts since birth as an inguinoscrotal swelling that extends to the abdomen. It may be missed in childhood and may present in adult life. Sac of ASH extends through the inguinal canal, and this part of the sac acts as an isthmus between the two sacs giving a dumbbell shape appearance.[2] However, our patient presented with predominant left-sided scrotal swelling and non-specific abdominal pain.

The abdominal sac can be retroperitoneal or preperitoneal. Sac is usually covered with the transversalis fascia. The abdominal sac may be of variable size, as large as a football, and may be of variable shape.[1] Although the scrotal sac is of varying size but is usually small. These sacs can be unilocular or multilocular. There is intercommunication between these two sacs without any communication with the peritoneal cavity.

There are various theories proposed for the development of ASH. Dupuytren had hypothesized that ASH occurs due to an extension of the scrotal sac into the abdomen through the inguinal canal whenever the scrotal sac is under high pressure.[1] Jacobson had hypothesized that ASH occurs as a result of an extension of infantile hydrocele.[4] Few others

added another concept to this hypothesis: the coexistence of inguinal defect and infantile hydrocele.[5] Roller hypothesized that encysted hydrocele of cord and ordinary hydrocele might form ASH; however, they did not explain the intercommunication between the sacs.[1] There are few other theories that have been proposed, but these have been less accepted. ASH can occur due to an extension of the preformed peritoneal sac extending into the scrotum through the inguinal canal.[1] ASH can occur due to the flow of fluid downwards through persistent processus vaginalis, which acts as a one-way valve.[6]

ASH occurring in the pediatric age group is known to have spontaneous regression, but such resolution has not been noted in adults. The patient presents with painless inguinoscrotal swelling and abdominal swelling. These swellings may be of variable size. Scrotal swelling may be reducible or may be partially emptied in the supine position. Cross fluctuation may be present due to the intercommunication of the two sacs, which is diagnostic for ASH. The presence of springing back ball sign and hourglass transillumination are diagnostic of ASH.[7] In our patient, there was the presence of cross fluctuation, but it was not transilluminating. This may be because of the brownish muddy fluid as the content due to hemorrhage in the cyst. Other associated anomalies are contralateral hydrocele, hernia, ectopic testis, cryptorchidism, and various urinary tract anomalies. In our patient, there was no associated congenital anomaly.

Long-standing ASH may have complications like testicular dysmorphism, hydronephrosis,[8] lymphedema,[9]pyocele, torsion, hematocele,[10] and rarely malignancy, which can be either testicular or paratesticular.[11]

The presence of hematocele or bleeding inside the sac can occur and should raise the suspicion of the existence of malignancy. It can occur as an acute event or maybe a chronic event. It may be discovered accidentally. Hematocele can occur due to spontaneous rupture, or trivial trauma, or malignancy.[10,11] Our patient had hematocele as the complication, probably developed since last two months as the patient presented with pain. Ultrasound also helps for diagnosis and can tell about the size and contents of the sac.

CT and MRI are additional investigations that can confirm the diagnosis and differentiate it from other diseases that mimic ASH. The presence of malignancy, associated congenital abnormality, or presence of testicular dysmorphism, etc. can be seen in CT and MRI. In our

case, there was a presence of testicular atrophy without any other congenital abnormality. The presence of hematocele may not be diagnosed preoperatively, even with an imaging technique. Many times hematocele will be diagnosed intraoperatively. In our case, imaging was not suggestive of hematocele and was showing testicular atrophy.

ASH can be treated with a conservative or surgical approach. A conservative approach with the expectant spontaneous resolution has been seen in asymptomatic and uncomplicated pediatric ASH; still, such an approach is not useful in adults as the spontaneous resolution does not occur in adults. Surgical procedures include excision of sac, aspiration or incision, and drainage.[1] Excision of the sac is the ideal treatment option, which can be done laparoscopically or by open approach. A laparoscopic approach was first tried in 2004. Boulhadiba et al. had performed the first successful laparoscopic ASH excision in 2007.[12]Excision of the abdominal sac along with partial excision of the scrotal sac with eversion most standard technique used. Sac can be excised en-bloc along with cord and testis whenever there is suspicion of testicular atrophy or testicular malignancy. Partial excision of the scrotal sac with drainage of the abdominal sac can also be done. Sac can be excised partially. In our patient, we could excise both sacs entirely with the removal of the left testis as there were testicular hematoma and atrophy.

Laparoscopy helps in confirming the diagnosis as well as used in definitive management. The presence of large ASH and for a long time may make the laparoscopic treatment difficult due to adhesion or proximity to a vital structure, scenario seen in our case. Open conversion may be needed in such cases. Clinicians should be cautious in patients dealing with large ASH as open conversion may be required. This case would give us worth attention towards the possibility of complicated ASH within, even if the preoperative imaging studies did not show any evidence of complications within ASH. Even in complicated ASH, we recommend laparoscopic treatment of ASH if the sacs can be excised safely. Also, there may be a concomitant hernia in patients with ASH or may develop in the future if the internal ring is dilated. Simultaneous repair of the dilated internal ring or hernia repair, if present, can be to prevent future occurrence of hernia.

Conclusion

ASH is a rare entity; although described mainly in the pediatric age group, it can also occur in adults. Clinical findings assisted by radiological imaging are diagnostic. It is usually

asymptomatic, and the development of symptoms is a sign of complicated ASH. Abdominoscrotal hematocele is a rare complication of ASH, and to the best of our literature search, it is the first case to document. Surgical excision is the definitive treatment. It can be done by open or laparoscopic approach. The presence of a concomitant hernia or weak abdominal wall should be repaired at the same time. If there is a presence of only a dilated internal ring, it can be repaired at index surgery.

References

1. Gadelkareem RA. Abdominoscrotal hydrocele: a systematic review and proposed clinical grading. Afr J Urol. 2018;24:83–92. https://doi.org/10.1016/j.afju.2018.01.006.
2. Ross JH, Kay R. Abdominoscrotal hydrocele in the infant. Clin Pediatr (Phila). 1992 May;31(5):317-9. doi: 10.1177/000992289203100514.
3. Mishra J, Behera TK, Rout B, Jena K, Ray Mohapatra CK. Hydrocele-en-bisac in a young adult: a rare encounter. ANZ J Surg. 2013 Jul;83:581-2. doi: 10.1111/ans.12220.
4. Cuervo JL, Ibarra H, Molina M. Abdominoscrotal hydrocele: its particular characteristics. J Pediatr Surg 2009;44:1766–70. DOI: 10.1016/j.jpedsurg.2008.12.002.
5. Singh D, Aga P, Goel A. Giant unilateral hydrocele "en-bisac" with right hydronephrosis in an adult: A rare entity. Indian J Urol2011;27:142-3. doi: 10.4103/0970-1591.78413
6. Celayir AC, Akyuz U, Ciftlik H, et al. A critical observation about the pathogenesis of abdominoscrotal hydroceles. J Pediatr Surg 2001;36: 1082-4.https://doi.org/10.1053/jpsu.2001.24760
7. Cozzi DA, Mele E, Ceccanti S, Pepino D, d'Ambrosio G, Cozzi F.Infantile abdominoscrotal hydrocele: a not so benign condition. J Urol.2008;180:2611–5.doi: 10.1016/j.juro.2008.08.054.
8. Avolio L, Chiari G, Caputo MA, Bragheri R. Abdominoscrotal hydrocele in childhood: is it really a rare entity?. Urology. 2000;56:1047-9. doi: 10.1016/s0090-4295(00)00801-3.

9. Belman AB.Abdominoscrotal hydrocele in infancy: a review and presentation of the scrotal approach for correction. J Urol 2001; 165: 225.https://doi.org/10.1097/00005392-200101000-00065
10. Estevao-Costa J, Morgado H, Soares-Oliveira M, Campos M, CarvalhoJL. Hemorrhagic abdominoscrotal hydrocele. A challenging entity. J Pediatr Surg. 2005;40:731–3.DOI: 10.1016/j.jpedsurg.2005.01.012
11. Bisceglia M, Dor DB, Carosi I, Vairo M, Pasquinelli G. Paratesticular mesothelioma. Report of a case with comprehensive review of literature. Adv AnatPathol. 2010;17:53-70. doi: 10.1097/PAP.0b013e3181c66fbc.
12. Bouhadiba N, Godbole P, Marven S. Laparoscopic excision of abdominoscrotal hydrocele. J Laparoendosc Adv Surg Tech A. 2007;17:701-3. doi: 10.1089/lap.2006.0249.

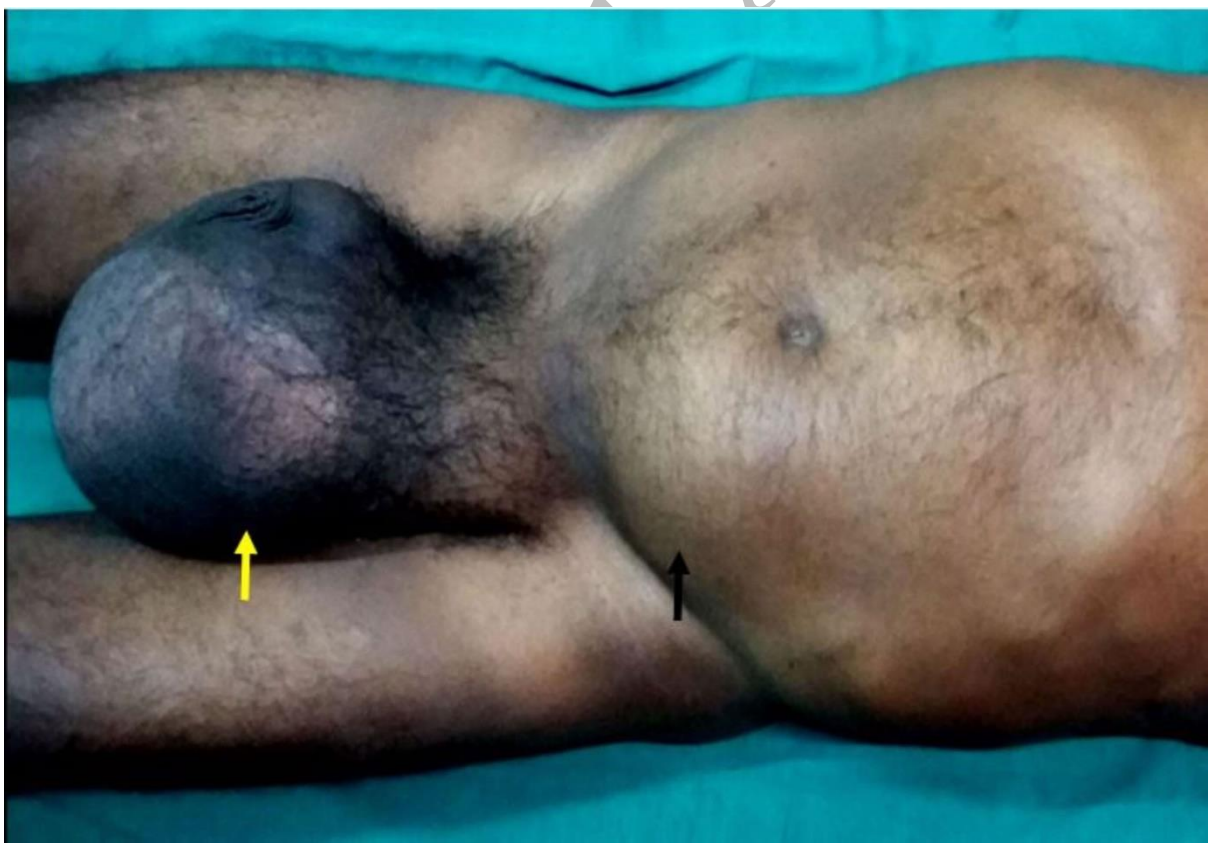

Figure 1: Clinical picture showing abdominal swelling (black arrow) and scrotal swelling (yellow arrow).

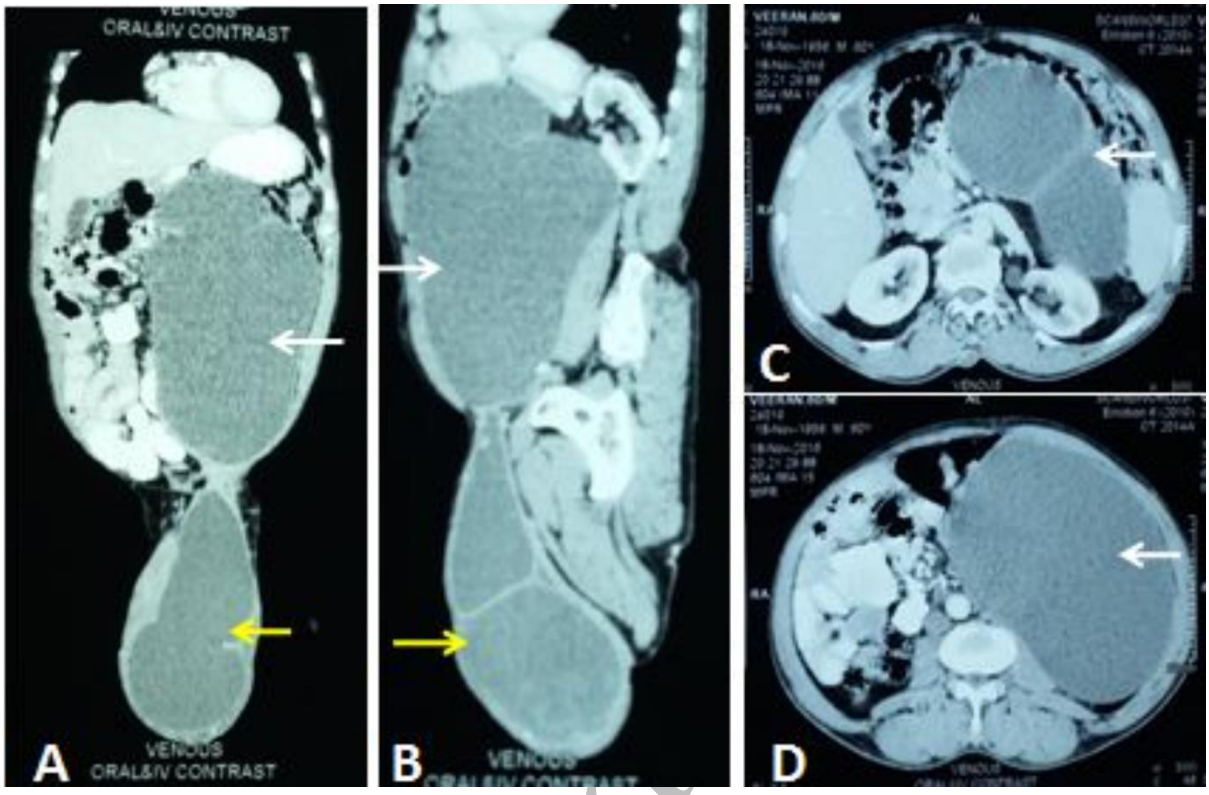

Figure 2: CECT images showing; Large hypointense walled collection 24 x 16 cm in the left side of the retroperitoneum (white arrow) extending in to the epsilateralinguino-scrotal region (yellow arrow) measuring 18 x 9 cm.

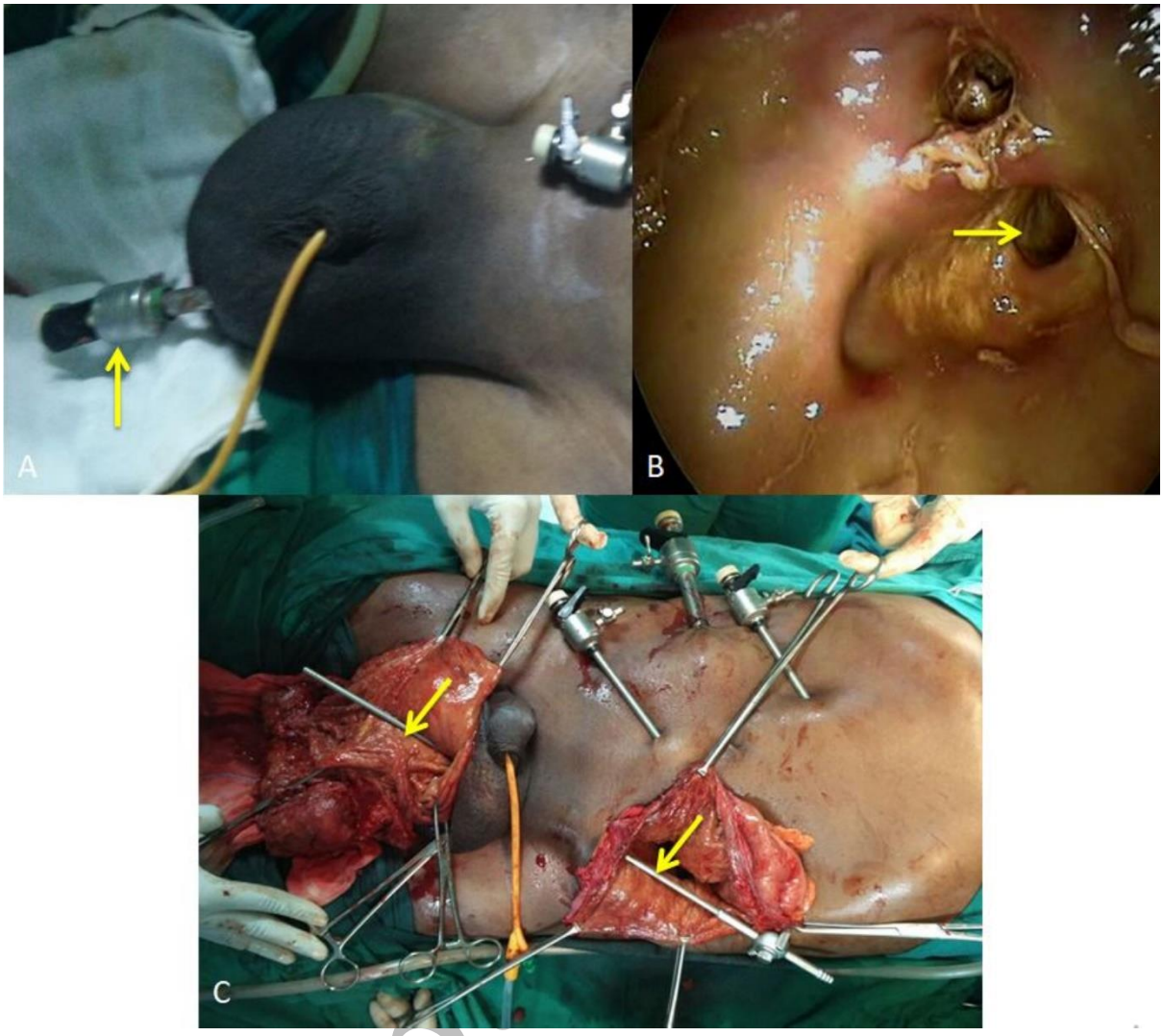

Figure 3: Intra operative images showing; **A:** Scrotal trocar (arrow) **B:** Trans-scrotal endoscopic view of scrotal sac depicting the opening of communication (arrow) with abdominal sac, and **C:** Communication between abdominal sac and scrotal sac by passage of laparoscopic suction cannula (arrows).

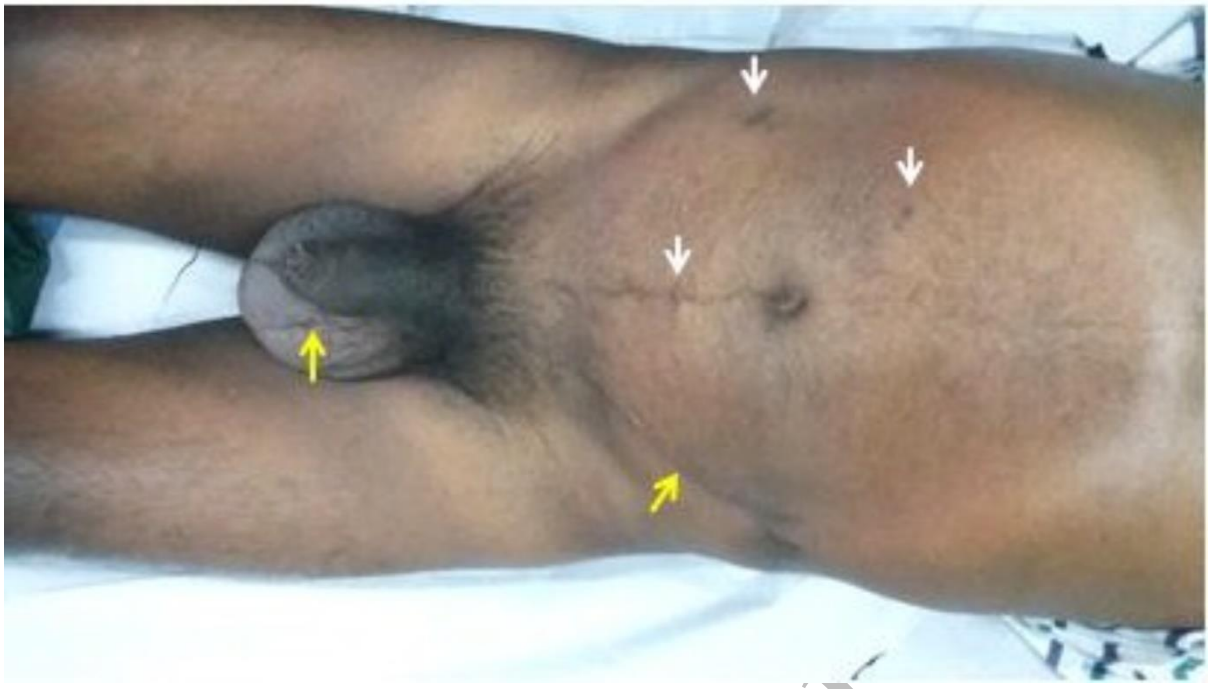

Figure 4: Clinical image during follow up of patient showing healed port sites (white arrows) inguinal and scrotal scars (yellow arrows).