CASE REPORT

Management of cavernous abdominal wall defects post radical cystectomy in adult exstrophy complex

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SUMMARY

The exstrophy–epispadias complex represents a spectrum of genitourinary malformations ranging from simple glanular epispadias to an overwhelming multisystem defect, cloacal exstrophy. Neonatal total reconstruction of bladder exstrophy–epispadias complex is the treatment of choice. An adult patient presenting with untreated exstrophy is very rare. Malignant transformation, commonly adenocarcinoma, in such cases is a known complication due to mucosal metaplasia of urothelium. Management in such cases necessitates a radical surgical procedure that often results in a massive defect in the anterior abdominal wall. Providing a cover for such defects is a challenging task for the reconstructive surgeon. Local skin flaps and wide mobilisation of the rectus muscle are the usually employed techniques for closure of such defects. However, these may be inadequate in extremely large defects such as those encountered in our patients. We, hereby, describe our technique of closure of the abdominal wall defect using a pedicled anterolateral thigh flap.

anteriorly. Bilateral testes were descended. Scrotum was well developed.

Second patient, an 18-year-old young woman, presented with a mass arising from exstrophic bladder for 2 months (figure 2). Vaginal introitus could not be identified separately. Bilateral inguinal lymph nodes were not palpable. Pubic diastasis was present with the anal opening situated anteriorly. Rectal mucosa was free from the mass.

INVESTIGATIONS

The haematological and biochemical parameters of both patients were within normal limits. Edge biopsy in both cases was suggestive of adenocarcinoma. CT scan of abdomen in first patient was suggestive of 7×6 cm heterogenous mass lesion involving the exstrophic bladder with enlargement of right external iliac and bilateral inguinal lymph nodes. CT abdomen of the second patient was suggestive of 8×7 cm heterogenous mass lesion involving the exstrophic bladder with no significant lymphadenopathy.

BACKGROUND

Although rare, malignant transformation of untreated exstrophy bladder in adults is a disastrous complication and a challenge for the treating surgeon. Wide resection to achieve negative margins results in wide abdominal wall defects that need to be covered with autologous tissue. We have elucidated in this report how skin from the anterolateral thigh (ALT) can be used to cover these defects, in a simple and consistent manner.

CASE PRESENTATION

Two adult patients presented to us with untreated exstrophy bladder. Both of these patients did not seek or receive any treatment in the past due to a combination of limitation of healthcare access, economic reasons and social stigma associated with such kind of genetic deformity.

First patient, a 48-year-old man, presented with discharge and bleeding from the protuberant bladder for 3 months. On examination, he had complete exstrophy–epispadias with cauliflower-like friable growth (6×3 cm) at right lateral margin of the bladder extending onto skin with adjacent induration about 1 cm beyond the visible margin (figure 1). We also noticed the presence of bilateral fixed hard inguinal lymph nodes. Pubic diastasis was present, with anal opening situated

TREATMENT

Surgery was carried out as a joint endeavour of a team comprising urologists and plastic surgeons. First patient underwent radical cystoprostatectomy including a 3 cm margin of the clinically normal skin with bilateral extended pelvic lymphadenectomy and bilateral radical ilioinguinal lymph node dissection with ileal conduit diversion. The resultant defect was 22×18 cm in the anterior abdominal wall (figure 3). As per preoperative plan, a flap including the skin, subcutaneous tissue and fascia lata was marked on the anterolateral aspect of the thigh. This flap was perfused by the perforators of the descending branch of the lateral circumflex femoral artery. It was raised as a pedicled flap and used to cover up the defect after being tunneled under the skin and subcutaneous tissue of inguinal region (figure 4). Fascia at donor site on right thigh was not approximated and closure was done using a split-thickness skin graft harvested from the left thigh to cover it.

Second patient underwent anterior exenteration and bilateral pelvic lymph node dissection. The anterior abdominal wall defect measured 20×15 cm and a pedicled ALT flap was raised and tunneled as described previously to cover up the defect. The donor site of the flap was covered similarly using split-thickness skin graft (figure 5).

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Figure 1 Preoperative clinical picture of index patient 1.

Duration of surgery was 9 hours and 8.5 hours, and blood loss was 600 mL and 700 mL, respectively.

OUTCOME AND FOLLOW-UP

First patient had an uneventful recovery except for superficial infection along left inguinal suture line (Clavien-Diindo I), which was managed conservatively. He was allowed liquids on postoperative day 3. Final histopathology was adenocarcinoma bladder with transmural infiltration up to adipose tissues; 3/5 right iliac lymph nodes were positive (pT3N2M0). Both ureteric ends, seminal vesicle and prostate were free. He was discharged on postoperative day 20. There were no graft-related complications (figure 6). He was started on Folinic Acid 5 Fluorouracil Oxaliplatin (FOLFOX)-based chemotherapy after 3 months following complete healing of surgical wounds.

Second patient was allowed orally on postoperative day 3. She developed superficial epidermal necrosis of the flap (Clavien-Dindo III b). On debridement, the underlying dermis was found to be healthy and vacuum-assisted closure was applied on sixth postoperative day. A split-thickness skin graft was used to cover the debrided healthy wound on postoperative day 16. No further complications were noted. The graft took up well as observed on

Figure 2 Preoperative clinical picture of index patient 2.

Figure 3 Intraoperative picture showing the cavernous abdominal wall defect in patient 1.

follow-up visits. Final histopathology of the specimen showed adenocarcinoma bladder with all margins and ureteric ends free. Separate tissue which was sent from anterior perirectal tissue with suspicion of tumour deposit came as positive. All lymph node packets were negative (pT3N0M0). She was discharged on postoperative day 25 (figure 7). FOLFOX was started after 3 months postoperatively.

Both cases tolerated treatment well and remained under our follow-up for 20 months and 16 months, respectively.

DISCUSSION

Extensive full-thickness abdominal wall defects commonly occur following tumour resection in cases of exstrophy of bladder with malignant transformation, more so than in an uncomplicated case of exstrophy, because an additional 2–3 cm margin of the surrounding uninvolved skin is excised when resecting the malignancy. Objective is to achieve an adequate clinical resection margin. Here, in this case, we followed wide resection similar to principles of management of colorectal adenocarcinoma.¹ Additionally, there were palpable inguinal lymph nodes in one case and underlying induration under the skin adjacent to the tumour raising concern of contiguous spread of adenocarcinoma into the integument and thereby necessitating wider resection margin. The goal in the reconstruction of large defects is to restore the

Figure 4 Pedicled anterolateral thigh flap used to cover the cavernous abdominal wall defect in patient 1.

Figure 5 Immediate postoperative picture patient 2.

anatomical as well as the functional integrity of the abdominal wall, thus protecting the exposed intra-abdominal contents and preventing future complications such as hernia formation. The flap to be used for covering the defect needs to have a good

Figure 6 Postoperative picture at 6 months patient 1.

Figure 7 Postoperative picture at 6 months patient 2.

vascularity and should be simple to perform with minimal complications. It should provide stable and durable coverage.

Several techniques ranging from rectus muscle mobilisation, use of meshes and local skin covers have been described for coverage of such defects. Xiong *et al* described the use of a customised titanium mesh to bridge the gap in the pubic bone.² They performed an onlay mesh repair along with a lower abdominal island skin flap to cover the 12×8 cm lower abdominal wall defect. They did not observe any complications and the mesh was functioning well at follow-up. Lauro *et al* have described closure of the infraumbilical defect with Cardiff repair with onlay mesh repair.³ Abdominal wall closure was done with a fasciocutaneous M-plasty. Such a repair, however, may not be feasible in extremely large defects with unavailability of adequate skin locally. Savalia *et al* reported managing the abdominal wall defect in a similar case using rectus abdominis rotation flap based on inferior epigastric artery.⁴ Such repairs that use the locally available skin or require mobilisation of the rectus may lead to further weakening of abdominal wall and ileal conduit-related complications.

Use of ALT flap for adult cases of bladder exstrophy has rarely been described.⁵ We find the ALT flap to be an ingenious, reliable method to cover large anterior abdominal wall defects. The harvest of the fascia lata along with the flap reliably restores the integrity of the abdominal wall. This precludes the use of mesh and its associated complications. Large flaps of 25×15 cm can be raised reliably. It has a long, constant vascular pedicle making its survival predictable. Rapid healing with flap cover facilitates early adjuvant therapies, if needed. Also, this flap does not influence the decision of urinary diversion as it avoids mobilisation/weakening of abdominal wall. The minimal donor site morbidity of the ALT flap is an additional advantage. We did not use Vicryl

mesh as both the patients had infected long standing fungating lesions with increased probability of mesh infection in postoperative period. Further, mesh would have led to wound contraction and in setting of a planned ileal conduit diversion ALT flap was better suited.

Learning points

- Untreated exstrophy of the bladder poses a risk for malignancy. Although such cases are rare, they pose a challenge for the treating surgeon.
- Surgical treatment in these cases results in enormous abdominal wall defect that needs a cover that will protect the intra-abdominal structures and reinstitute the integrity of the abdominal wall.
- The pedicled anterolateral thigh flap has many benefits: provides a well vascularised cover; restores integrity of the abdominal wall; protects intra-abdominal contents; does not impact the decision of urinary diversion; facilitates early start of adjuvant therapies as there is rapid healing.
- Prognosis may be poor but proactive multimodality care in such cases has the potential of satisfying outcomes.

Contributors This particular case and gargantuan challenges posed during the management were dealt by joint endeavours of urology and plastic surgery department. First patient was managed primarily under RN and PK with inputs from

plastic surgery department represented by DG. The authors had discussion with plastic surgeons regarding the best possible way to manage such a challenge after resection of the tumour. Similarly, second patient was managed primarily under AS in collaboration with plastic surgeons and management was very much similar to first case as illustrated. Plastic surgeon, DG, explained to us in detail the various options available to us for abdominal wall reconstruction and guided us to the best possible option available with careful regard to the outcome. Surgery could be seen as consisting of two major steps; radical cystectomy was first done by urologists and plastic surgeon then took over the reconstructive surgery aided by urologists. All the authors were involved in drafting the initial manuscript and thereafter taking it to final version which is being submitted here, after scrutinising every minor details.

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