CASE REPORT

Recurrent hospitalisations in a rare case of hemicorporectomy: a challenging case for medical management

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SUMMARY

Hemicorporectomy, or translumbar amputation, is a radical surgery involving the dissection of the body at the waist and is usually reserved for complex medical conditions including locally invasive malignancy and terminal pelvic osteomyelitis. Only 71 cases have previously been reported. We present a rare case of hemicorporectomy in a 53-year-old patient with terminal pelvic osteomyelitis which occurred after he suffered a gunshot wound at T6 causing paraplegia at the age of 31. Unfortunately, this patient continued to suffer recurrent hospitalisations and sepsis events secondary to chronic, non-healing advanced pressure ulcers and complicated urinary tract infections despite repeated courses of broad-spectrum intravenous antibiotics and surgical debridements. In light of his diminished quality of life and poor prognosis, the patient's family chose to manage his condition conservatively with home hospice. This case illustrates the significant challenges in the medical and surgical management of hemicorporectomy patients.

BACKGROUND

Hemicorporectomy, or translumbar amputation, is a radical surgery involving dissection of the body at the waist (commonly between L4 and L5). It entails removal of the legs, genitalia, bladder, pelvic bones, anus and rectum, and is commonly performed in two stages, with a colostomy and ileal conduit being created during the first operation and the actual hemicorporectomy being done during the second operation. The first ever hemicorporectomy was performed in 1950, and as of 2017 a total of only 71 cases have been reported.^{1,2}

The original indication for hemicorporectomy was locally invasive malignancy of the pelvis for which chemotherapy, radiotherapy or conventional surgeries were not beneficial or possible. In recent years, it has been performed for other indications, including terminal pelvic osteomyelitis, squamous cell carcinoma (SCC) in a pressure ulcer, pelvic chondrosarcoma, severe pelvic trauma, vascular malformations, and in one case gangrene secondary to acute aortic occlusion.³ Of the 71 reported cases of hemicorporectomy performed, 21 (30%) cases have been reported for terminal pelvic osteomyelitis, 43 (60%) cases for malignancy (13 cases were SCC arising from pressure ulcers), 4 cases for

benign disease and 3 cases for trauma. Outcomes are generally more favourable for non-malignant versus malignant indications.⁴ For terminal pelvic osteomyelitis, more than 50% of patients survived at least 9 years, with an average survival of 11 years posthemicorporectomy.⁵ The average survival post-procedure decreases to 2.9 years when malignancy is the indication for hemicorporectomy.⁵ Overall, one-third of patients survived at least 9 years after hemicorporectomy.⁵

Although published reports suggest prolonged survival (>50% survival for terminal pelvic osteomyelitis at 9 years) posthemicorporectomy, this does not take into account the quality of life and morbidity of patients.⁵ In the postoperative period, complications of hemicorporectomy include volume overload, wound dehiscence and delayed healing.⁵ Some of the first patients to undergo this procedure died of pulmonary hypertension.⁶ For patients with long-term survival, other factors to consider include decreased body mass and circulating blood volume, as well as loss of muscle mass, which is thought to affect fluid, acid-base, electrolyte balance and drug dosing.⁷ Moreover, the radical nature of a hemicorporectomy severely distorts the body anatomy and may introduce new problems such as faecal and urine drainage from ostomy sites into dependent areas, with chronic non-healing pressure ulcers causing infections and complicated urinary tract infections (UTIs) resulting in repeated sepsis events requiring multiple hospitalisations. Furthermore, it has also been reported that the loss of body surface area also affects heat dissipation and temperature regulation.⁸ Taken together, these factors can potentially diminish the quality of life and increase the morbidity of hemicorporectomy patients, which calls into question the best way to manage the complications of hemicorporectomy patients and provide comprehensive care in the inpatient and outpatient settings.

CASE PRESENTATION

Here, we present a 53-year-old African-American man who underwent a hemicorporectomy at the level of L2 (at age 49) after a series of surgical procedures for terminal pelvic osteomyelitis, chronic advanced pressure ulcers and chronic complicated UTIs, which occurred after a gunshot wound at T6 causing paraplegia at the age of 31 (box 1, figure 1).

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Box 1 Timeline of significant medical and surgical events

- ▶ 1994: initially became paraplegic secondary to a gunshot wound at T6.
- ▶ July 2006: elective transverse loop colostomy with subsequent excision and creation of ileostomy.
- ▶ November 2007: right hip disarticulation and amputation due to right femoral head osteomyelitis.
- ▶ December 2007: stage 4 sacral pressure ulcer, left second toe osteomyelitis.
- ▶ January 2008: recurrent urinary tract infections (UTIs) secondary to indwelling Foley catheter.
- ▶ February 2008: suprapubic catheter placed secondary to paraphimosis and erosion of the urethral meatus secondary to the indwelling Foley catheter.
- ▶ April 2008: sepsis secondary to penile shaft necrosis and penile ulcers; suprapubic catheter malfunction, with subsequent sepsis secondary to UTI from urocutaneous fistula.
- ▶ December 2009: ileovesicostomy using ileum as a loop and primary bowel anastomosis.
- ▶ 2011/2012: urosepsis, stage 4 chronic sacral pressure ulcer debridement, septic left hip and sacral bone biopsy consistent with osteomyelitis.
- ▶ April 2012: multiple left lower extremity pressure ulcers with necrotic left toes; subsequent left hip disarticulation and translumbar amputation at L2 leading to his current state: hemicorporectomy.
- ▶ September 2012: enlargement of bilateral back and hip pressure ulcers; excisional debridement.
- ▶ November 2013: revision of colostomy due to prolapse; cystectomy and excision of ileovesicostomy; ileal conduit urinary diversion and bilateral ureteral stent placement.
- ▶ January 2016: prolapsed ileal loop, requiring revised ostomy.
- ▶ September 2016: sepsis due to large chronic back pressure ulcer—50×50 cm, grades 1–4.
- ▶ October 2016: patient dies in hospice care: sepsis secondary to advanced pressure ulcers.

Figure 1 Hemicorporectomy patient showing (A) urostomy with ileal conduit and (B) ileostomy.

His medical and surgical history was significant for hemicorporectomy with colectomy, splenectomy and creation of ileostomy and urostomy with ileal loop, multiple infected posterior and inferior thoracic pressure ulcers of all stages status post multiple debridements, terminal pelvic osteomyelitis, recurrent multi-drug-resistant complicated UTIs with sepsis, methicillin-resistant *Staphylococcus aureus* (MRSA) and vancomycin-resistant *Enterococcus* (VRE) bacteraemia with sepsis, hypertension, anaemia of chronic disease, chronic kidney disease (CKD), gastritis, oesophagitis, and pulmonary embolism. He presented to the Sinai-Grace Hospital Emergency Department with fever (reported by caregiver to be >39°C at home) and altered mental status. On admission, he was found to be hypotensive (systolic blood pressure ~82 mm Hg), with leukocytosis (white blood count (WBC) $14.1 \times 10^9/L$ with neutrophilic predominance and bands of $1.6 \times 10^9/L$) and acute kidney injury on CKD (creatinine 1.36), and was diagnosed with severe sepsis likely secondary to an infected advanced chronic non-healing posterior thoracic pressure ulcer and a possible complicated UTI. He was intubated and mechanically ventilated in the medical intensive care unit (MICU) for airway protection due to his altered mental status (alert and oriented $\times 0$). He was initiated on empiric antibiotics including intravenous cefepime and clindamycin, along with fluid resuscitation. Infectious disease was consulted for antibiotics management due to the patient's history of multi-drug-resistant organisms (MDROs). However, blood cultures remained negative during this admission. Urinalysis was positive for nitrite and leukocyte esterase, with >100/high power field (HPF) urine WBCs. Lactic acid was 1.7 on admission. Urine culture grew >100 000 colony-forming units/mL with multiple organisms present.

The patient's mentation improved (alert and oriented $\times 3$), and he was subsequently extubated on admission day 1. General surgery was consulted for surgical debridement of his extensive, advanced posterior thoracic pressure ulcer spanning 50 cm $\times$ 50 cm at various stages (1–4) of healing (figure 2). All necrotic tissue was excised to obtain viable bleeding tissue and haemostasis was achieved. Appropriate postoperative wound care was initiated with daily dressing changes with wet-to-dry dressings, soft care mattress, every 2-hour turns and adequate pain control. His ileostomy bag was replaced to ensure a proper seal to prevent faecal drainage into his posterior thoracic pressure ulcer. In addition, nutrition was consulted to optimise his nutritional intake to support wound healing. Antibiotics were continued for 7 days as per infectious disease recommendations. Our patient was discharged home on admission day 7. He lived with his sister, who was his caregiver and power of attorney. Appropriate postdischarge wound care was initiated with home healthcare services for daily dressing changes, nursing care for ileostomy and urostomy changes, and home physical therapy. In addition, follow-up with the wound care clinic was arranged.

INVESTIGATIONS

See the Case presentation section.

TREATMENT

See the Case presentation section.

OUTCOME AND FOLLOW-UP

Our patient was readmitted 3 weeks later for another episode of sepsis with altered mental status. He was appropriately treated and recovered to his baseline. Psychiatry was consulted, and it was determined that he was not competent to make decisions

Figure 2 Evolution of pressure ulcers. (A) September 2011: stage 4 sacral pressure ulcer 7×9.5×2.8 cm, (B) January 2013: stage 3 left thorax pressure ulcer 31.5×11×0.1 cm, and (C) September 2016: stages 1–4 posterior thorax pressure ulcer 50×50 cm.

regarding his medical care. In light of the fact that this patient had been admitted to the hospital on average 10 times/year in recent years, with 11 admissions over the last year alone, palliative care was consulted to assess patient and family wishes and determine goals of care. After an extensive discussion with the patient and his family regarding his diminished quality of life and poor prognosis, his sister, the durable power of attorney, decided that it was best for him to enrol in home hospice, with a focus on comfort care. The patient was subsequently admitted to inpatient palliative care 2 weeks later with altered mental status. He subsequently died during this admission in palliative care.

This case highlights the significant medical, surgical, financial and social challenges faced by the families, their caregivers and medical professionals involved in the care of hemicorporectomy patients.

DISCUSSION

Here we present a case report of a 53-year-old African-American man who suffered a gunshot wound at T6 causing paraplegia at the age of 31 and subsequently underwent a hemicorporectomy at the level of L2 at the age of 49 for terminal pelvic osteomyelitis in a series of body-preserving surgical procedures over a period of 5 years (box 1). This radical surgical procedure prolonged his life, but arguably did not reduce his morbidity and significantly distorted his body anatomy, which presented unique challenges in his medical care. The close proximity of his anterior thoracic urostomy and ileostomy to each other and to his pressure ulcer frequently lead to stool and urine draining into his chronic pressure ulcer located on his posterior and inferior thorax during ileostomy and urostomy bag changes (figure 1). As a result, he required recurrent hospitalisations for sepsis secondary to complicated UTIs and chronic, non-healing advanced pressure ulcers, which gradually progressed and evolved to encompass larger areas of his posterior and inferior thorax, further altering his body morphology (figure 2). Surgeons performing repeated pressure ulcer debridement frequently had difficulty describing the exact location of the ulcer due to his distorted body anatomy. Furthermore, our patient had repeated ostomy prolapses requiring multiple surgical revisions likely due to the high intraperitoneal pressures generated by his body weight on peritoneum without a supporting bony frame (figure 2, box 1).

During each of his numerous hospital admissions, healthcare professionals struggled with his presenting diagnosis and the management of his multidrug-resistant infections, electrolyte abnormalities, monitoring kidney function and blood pressure control. Infectious disease was frequently consulted to manage this patient due to his history of MDROs, and he was invariably placed on multiple broad-spectrum antibiotics. A biopsy of his chronic advanced non-healing pressure ulcer grew *Acinetobacter baumannii* resistant to all major classes of antibiotics (including carbapenems), MRSA and Zosyn-resistant *Pseudomonas aeruginosa*. He also had numerous bouts of VRE and MRSA bacteraemia, which required alternate broad-spectrum antibiotics such as daptomycin for treatment. In addition to

increasing the cost of his medical care, repeated hospitalisations and treatments with broad-spectrum antibiotics also increased the chances of MDRO dissemination to other patients in the hospital.

This patient also required unconventional drug dosing due to his altered body composition and decreased body weight. For example, our patient had a total body weight of 30 kg with a reduced proportion of muscle mass since he was lacking his pelvis, sacrum and bilateral lower extremities. Therefore, his antibiotics, antihypertensive regimen and venous thromboembolism prophylaxis regimen had to be altered to adjust for his body mass and composition. His altered anatomy (urostomy located beside ileostomy) and body composition also presented diagnostic challenges, including persistent pyuria with positive nitrite and leucocyte esterase, which made it difficult to determine if his recurrent sepsis events were attributed to a chronic complicated UTI or his chronic, non-healing advanced pressure ulcers. In addition, he had a baseline systolic blood pressure between 90 and 100 mm Hg, which is not uncommon in patients with hemicorporectomy and paraplegia. In the setting of sepsis, a small reduction in blood pressure when combined with errors in measurement can result in a false diagnosis of septic shock, which in this case frequently prompted placement of a central line and MICU admission as a precautionary measure. These factors undoubtedly increased patient discomfort, length of hospital stay and significantly increased cost burden to the healthcare system.

This case also presented unique challenges for allied health professionals, including nurses and social workers. This patient required long-term specialised urostomy and ileostomy care, as well as wound care and daily dressing changes for his chronic non-healing pressure ulcers. Appropriate outpatient follow-up, education and training for caregivers on aseptic technique and patient hygiene are crucial to decreasing mortality and morbidity and improving the quality of life for hemicorporectomy patients.

The management of chronic non-healing advanced pressure ulcers is generally very costly to the healthcare system and significantly reduces the quality of life of the patient. Alternative treatment options such as maggot debridement therapy and hyperbaric oxygen therapy are currently in development and have shown promising results in institutions that have access to such treatments.^{9 10} More research examining the efficacy and cost-effectiveness of various prevention and treatment strategies is needed, ideally with the goal of establishing specific guidelines.

This case also highlights the need to educate the patient and family and establish goals of care in hemicorporectomy patients, with quality of life and patient wishes as the guiding principles. A patient's mental well-being including decision-making capacity, self-image, ability to perform activities of daily living, the potential desire to return to work or school, financial concerns, potential social isolation, pain management and radical changes in ability to perform specific sexual activities must also be addressed. The patient involved in this case had clearly expressed his wish to live and share his life with his son

and family until late 2016, when his disease burden overtook his ability to make rational decisions, requiring a durable power of attorney for decision-making.

Although the patient in this case provides a unique account of the significant healthcare challenges in managing the potential complications of some hemicorporectomy patients, it is important to highlight that many patients do enjoy a productive life and experience a less complicated posthemicorporectomy course. For example, Peterson and Sardi¹¹ describe a case of hemicorporectomy for SCC (Marjolin's ulcer), where the patient remained disease-free for at least 7 years (as of November 2003) after hemicorporectomy and was employed as a computer programmer, independent in activities of daily living and retained the ability to drive. Other case reports suggest that some hemicorporectomy patients can live multiple decades despite a grave preoperative diagnosis.¹² A case series of one woman and eight men (all paraplegic) who underwent hemicorporectomy for terminal pelvic osteomyelitis between 1981 and 2005 found that for the four surviving patients at follow-up, none suffered recurrent decubitus ulcers (with a range of survival after surgery of 1.7–22 years).⁵ For the patients who died, the cause of death was urosepsis for one patient, a fall for one patient, portal vein thrombosis for one patient and unknown cause for two patients. This makes our patient unique as the majority of posthemicorporectomy patients do not suffer recurrent osteomyelitis and advanced pressure ulcers leading to recurrent sepsis events. However, similar to our patient in this case, these patients had also undergone multiple surgical procedures including wound debridements and local flaps in an attempt to treat chronic pressure ulcers prior to hemicorporectomy. At the time that hemicorporectomy was considered, one patient had an ileostomy, four patients had colostomies and three patients had undergone procedures for urinary diversion.⁵

These notable examples illustrate that a significant number of hemicorporectomy patients can prolong their life as well as enjoy a relatively good quality of life with a multidisciplinary approach to care. After hemicorporectomy, all patients should undertake a rehabilitation process that involves the integration of different services such as occupational and physical therapy for increasing upper body strength and acquiring new mobility skills, nutrition, enterostomal therapy and wound care nursing, and psychological counselling.⁵ A previous study showed that patients who survived the procedure for terminal pelvic osteomyelitis and had long-term follow-up were likely to be happy with their decision to have had the procedure as it prolonged their lives and also relieved them from the pain of living with chronic pressure ulcers.⁵ Overall the procedure has both benefits and risks, and patients and their families should properly understand both implications and make the best decision while consulting with healthcare providers.¹³ Furthermore, Jankowski and Campo-Engelstein¹³ suggest that beyond assessing the risks and benefits of hemicorporectomy, the concept of informed consent, justice and resource allocation, and the loss and lived body are important considerations and remain topics of debate.

Unfortunately, there are currently no guidelines or standards of care for the management of hemicorporectomy patients, either on an inpatient or outpatient basis. This is primarily due to the rarity of the procedure and the paucity of subsequent data regarding outcomes, complications and treatments. We hope that this case may help healthcare providers anticipate and manage some of the potential complications in similar patients who have undergone hemicorporectomy or other radical surgical procedures.

Patient's perspective

The patient died in September 2016 after a recurrent bout of sepsis while he was in home hospice.

Learning points

- ▶ Hemicorporectomy patients require a multidisciplinary approach to care.
- ▶ Hemicorporectomy secondary to terminal pelvic osteomyelitis has the potential of prolonging life, but may also lead to a significantly diminished quality of life if adequate multidisciplinary care is not provided to the patient.
- ▶ Hemicorporectomy patients present unique challenges for inpatient and outpatient management.
- ▶ Early patient and family education regarding the quality of life and prognosis is required to establish goals of care for decision-making and provide options for comprehensive outpatient care including home healthcare services, physical and occupational therapy, psychiatric care, wound care, and even palliative/hospice care in select patients.

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Competing interests None declared.

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