C loacal exstrophy (CE) is a rare congenital complex deformity that is associated with anterior abdominal wall defects, reflex and exposure of the cloaca (uninterrupted ureter and bowel), aproctia, a widely separated pubic bone, and defects or dysplasia of external genital organs. Here, we present the case of a 42-year-old man with squamous cell carcinoma arising from an abdominal wall defect complicated by cloacal exstrophy. He was successfully treated with excision of the skin lesion with the bowel and reconstruction using a pedicled anterolateral thigh flap combined with a tensor fasciae latae flap. To our knowledge, this is the first report of squamous cell carcinoma arising from an abdominal wall defect complicated by cloacal exstrophy. (Plast Reconstr Surg Glob Open 2015;3:e315; doi: 10.1097/GOX.0000000000000286; Published online 2 March 2015)

CASE REPORT

A 42-year-old man was referred to our hospital with refractory skin ulcer in the lower abdominal region, an abdominal wall defect associated with CE, and stool-like discharge with straining. He had a 3-year history of repeated erosions or ulcers on the lesion. Soon after birth, a colostomy had been created on the left side of the abdomen and a cutaneous ureterostomy on the right side, and left nephrectomy had been performed. Pelvic osteotomy was not performed for a separated pubic bone. The patient had required hemodialysis for the last 18 years because of renal failure due to chronic pyelonephritis.

Physical examination showed a scar-like skin lesion of 9 × 12 cm, in which a hypopigmented lesion was combined with pigmentation and a 3 × 4 cm skin ulcer on the right side of the lesion, which was penetrating into the small intestine. The lesion caused abdominal wall hernia. Biopsy of the ulcer revealed SCC. Three weeks after biopsy, total resection of the scar-like skin lesion with the bowel was performed. At the time of the operation, the ulcer had rapidly increased in size to about 6 × 9 cm (Fig. 1). Defects of the penis and left testis were also seen. Computed tomography showed a widely separated

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The scar-like lesion resulted from a deficit of subcutaneous adipose tissue and muscle. Arteries in the abdomen and lower extremities were highly calcified due to long-term hemodialysis.

The total lesion of scar-like skin with a 1-cm margin was resected en bloc with about 25 cm of intestine involving an enterocutaneous fistula. The full-thickness abdominal wall defect was 11 × 15 cm in size. A 13.5 × 20 cm pedicled anterolateral thigh (ALT) flap with fascia latae and iliotibial tract was first designed in the patient’s left thigh. To obtain a large rotation arc and sufficient blood supply in the distal portion of the flap, distal perforators of descending branches of the lateral circumflex femoral artery are favorable. Furthermore, because of the separated pubic bone, the distance from the pivot point to the defect was a little longer than usual. However, a dominant distal perforator from the descending branch was not identified; only a dominant proximal perforator from the transverse branch was present. To gain as much blood supply as possible to the distal portion, a pedicled ALT flap combined with a tensor fasciae latae flap with a double vascular pedicle was finally harvested. The flap was transferred to the defect under the rectus femoris, sartorius muscle, and the subcutaneous space of the left lower abdomen superior to the left separated pubic bone.

In fluorescein angiography with indocyanine green (ICG), part of the distal portion of the flap did not show ICG, despite a width of only 2–3 cm, and was removed (Fig. 3). The flap was sutured firmly to the defect, nearly all-around except where the vascular pedicle passed through. The nonfirmly sutured part was considered fragile due to abdominal pressure. Therefore, the proximal portion of the skin paddle of the flap was de-epithelialized and sutured into the subcutaneous space to prevent herniation as far as possible. Alloplastic materials were not used because of suspected infection of the ulcer and bowel resection. The donor site was covered with a meshed skin graft from the right thigh.

A pathological examination revealed well-differentiated SCC in a white lesion and poorly differentiated SCC in an ashen lesion. The scar-like lesion differed from true scar tissue, but fibrosis, inflammatory cell infiltration, and edema were seen, unlike in normal dermal tissue (Fig. 4). Pathologically, the margin of the resected tissue was negative for SCC. The postoperative course was uneventful, and the wound healed without complications. The flap survived without congestion and ischemia. No apparent abdominal wall hernia was present 3 months postoperatively (Fig. 4).

**DISCUSSION**

CE is the most complex pathology of extrophy anomalies. The incident of this anomaly is 1 in 200,000 to 400,000 births and may be more common in males. The first successful reconstruction was reported by Rickham in 1960. In our case, pelvic osteotomy had not been performed for a separated pubic bone, although in neonates aged less than 48–72 hours, a single approximation of the pubic symphysis might help abdominal closure because the bones are still malleable, and no osteotomy may be required. Abdominal wall closure had also not been performed, resulting in abdominal wall hernia covered only with thin fragile scar-like skin.

Development of SCCs (Marjolin ulcers) is a late complication of burn scar, chronic venous insufficiency ulcers, vaccination scar, skin graft donor site, and gunshot wounds. The pathogenesis of malignancy in chronic scar is unknown. Research on burn-scar carcinoma has shown that these scars have...
decreased resistance to infection, are poorly vascularized, and are likely to ulcerate because of poor nutrition. It has been theorized that released tissue toxins may function as carcinogens or that such changes may make the scar more susceptible to damaging effects of ultraviolet radiation. In our case, the patient had a 3-year history of repeated erosions or ulcers, and the chronic abdominal wall defect may have been a cause of SCC. To our knowledge, this is the first report of SCC arising from an abdominal wall defect in a case with CE.

Reconstruction of defects of the abdominal wall after resection of a malignant tumor is often challenging because the defect can be large and the surgical field may be contaminated due to bowel resection. Therefore, alloplastic materials might be at risk for infection and exposure. For this reason, we used a flap with tensor fasciae latae and the iliotibial tract and deposited a de-epithelialized proximal skin paddle of the flap above the point at which the vascular pedicle passed through to prevent herniation, instead of using alloplastic materials.

The advantages of the pedicled ALT flap are that the blood supply to its distal portion is reliable and that the vascular pedicle is longer, if a dominant distal perforator is present, than that in a tensor fasciae
latae musculocutaneous flap, which has also been used for abdominal wall reconstruction. In our case, only a proximal perforator was identified, and therefore, a pedicled ALT flap combined with a tensor fasciae latae flap with a double vascular pedicle was harvested to provide as much blood supply as possible to the distal portion of the flap. ICG angiography is useful for intraoperative flap evaluation, and we found that the ICG did not dye part of the distal portion, despite this being only 2–3 cm in width. Fortunately, after removing the portion, the flap was transferred to the defect and sutured without pedicle tension. If the pedicle length has been insufficient, free ALT flaps may have been needed, despite the high risk of anastomosis thrombosis because of severe vessel calcification due to long-term hemodialysis.

CONCLUSION

In conclusion, a middle-aged man with SCC arising from an abdominal wall defect complicated by CE was successfully treated with excision of a skin lesion with the bowel and reconstruction using a pedicled ALT flap combined with a tensor fasciae latae flap.

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