



Case report

Case Report: Biologic graft placement with subsequent radiation therapy following radical vulvectomy for adenoid cystic carcinoma of the Bartholin's gland

Keely Ulmer^{a,*}, Megan E. McDonald^a, Joseph T. Kowalski^b

^a Department of Obstetrics and Gynecology, Division of Gynecologic Oncology, University of Iowa Hospitals and Clinics, Iowa City, IA 52242, United States

^b Department of Obstetrics and Gynecology, Division of Urogynecology, University of Iowa Hospitals and Clinics, Iowa City, IA 52242, United States

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ABSTRACT

Adenoid cystic carcinoma (ACC) of the Bartholins gland, first described by Klob in 1864, is a rare form of vulvar cancer comprising approximately 2–7% of all invasive vulvar lesions (Cardosi, 2001). Treatment consists of excision followed by radiation therapy (Cardosi, 2001; Anaf, 1999; Barcellini, 2020). Progression is indolent with later recurrence and metastases in comparison to other forms of vulvar cancer (Yang, 2006). Resection remains the gold standard for treatment followed by radiation therapy if margins are positive (Cardosi, 2001; Yang, 2006; Chang et al., 2019). We present a case of ACC of the Bartholins gland that underwent radical vulvectomy and Surgisis graft placement due to the extent of disease resection. Radiation therapy was then pursued due to positive margins with no wound breakdown despite this being the most common complication of vulvectomy with or without radiation therapy (Leminen et al., 2000). To our knowledge this is only the second case of Cook Biodesign graft placement after vulvectomy and first case of subsequent local radiation therapy to the area.

1. Case report

A 67-year-old female was referred to gynecologic oncology after diagnosis of ACC of the Bartholin's gland. The patient experienced a few months of vulvar discomfort and small amount of bleeding with wiping. She was seen by her primary provider who noted a vulvar lesion and referred her to gynecology. Exam under anesthesia was performed which revealed a left vaginal lesion. Biopsy confirmed the diagnosis. After presentation to gynecologic oncology clinic pelvic exam revealed a 5x6 cm mass centered around the left Bartholin's gland. The mass was felt to be separate from the rectal mucosa and urethra, it was encroaching upon the left urethral meatus by approximately 1 cm but felt to be without invasion of the pubic rami. Decision was made to proceed with surgical resection and possible skin flap or muscle closure pending Positron Emission Tomography/Computed Tomography further characterization. PET/CT revealed mild to moderately hypermetabolic left vulvar tumor involving the left vaginal wall without evidence of lymphadenopathy or distant metastases. MRI was then performed which described an enhancing mass involving distal left vagina, left introitus, left labia, and left perineum. The mass invaded the left puborectalis and inferior left levator ani muscles. The mass also abutted and displaced vagina, rectum, and urethra without evidence of

metastases.

The patient was taken to the operating room where the mass was dissected from the urethral meatus and rectum and radical vulvectomy performed. The majority of the defect was closed primarily, however a 4x3 cm defect remained in the vulva. Decision was made to proceed with closure using a Cook Biodesign 4-ply graft with completely tension free application. This was approximated with interrupted 2–0 Vicryl sutures around the periphery and in the mid portion of the graft to attach to underlying subcutaneous tissues. A specialist in Female Pelvic Medicine and Reconstructive Surgery assisted with the placement of the biologic graft given their experience with the usage of grafts in both gynecologic prolapse repair and for recurrent mesh exposure/erosions with poor wound healing/re-epithelialization of the tissues. The patient was seen two weeks post operatively with well healing post-operative wounds. Surgical pathology returned as ACC with positive margins as expected. She then completed 5580 cGy to the pelvis and vulva. Five months post operatively the graft was noted to have completely epithelized without evidence of disease recurrence. She is currently undergoing routine clinical surveillance and is without wound complication or graft breakdown seven months post operatively.

* Corresponding author at: 200 Hawkins Drive, 31506 PFP, Iowa City, IA 52242, 319-356-2016, United States.

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2. Discussion

ACC of the Bartholins gland is a rare entity requiring surgical excision and possibly radiation therapy (Cardosi, 2001; Anaf, 1999; Barcellini, 2020; Yang, 2006; Chang et al., 2019). Only one prior case report demonstrated the use of a biologic graft after vulvectomy. In the case described by Bratila et al, radical vulvectomy was performed for recurrent verrucous carcinoma of the vulva requiring multiple resections. This case described the use of Surgisis porcine graft with favorable patient outcome and healing of the wound defect (Brătîlă, 2015). The other uses for these types of grafts in the perineum or vagina found in our literature search are for urogynecologic applications such as recurrent mesh erosions/exposure and two case reports of vaginoplasty for Mullerian agenesis (Ding, 2013; Jakus et al., 2008). Additionally, an observational study conducted by colorectal surgery examined long term outcomes of biologic mesh in rectal cancer following extralevator abdominoperineal excision of rectum (ELAPER) (Thomas, 2019). To our knowledge, our case is the only case in the literature of biologic graft placement which was followed by radiation therapy to the area for the treatment of ACC. Fig. 1.Fig. 2.

The type of biologic graft used was the Cook Biodesign 4-ply graft which is an acellular matrix biomaterial harvested from porcine small intestinal submucosa. This graft contains critical components considered key in supporting granulation and epithelization in wounds and providing a scaffold for tissue repair such as collagen, proteoglycans, and growth factors. It is commercially available in biological dressings designed to mimic the human cutaneous natural extracellular matrix and initially received regulatory clearance in August 1997 and is currently available in the United States, Canada, Mexico, Europe, and Australia (Nherera, 2017). There is a paucity of data regarding the use of biologic grafts in vulvar pathology. In the study done by colorectal surgery, comparing myofascial flaps to biologic grafts following excision of the rectum due to anorectal cancer there was no difference in the rates of complications namely poor wound healing, perineal herniation, and



Fig. 1. Photo of Cook Biodesign 4-ply graft placed after radical vulvectomy.



Fig. 2. Vulvar wound prior to radiation.

chronic perineal pain. Of interest is the fact that these patients undergo extended duration preoperative chemoradiation to the area prior to this surgery. After the placement of these grafts the patients did not undergo any graft specific follow up and were followed routinely. The authors also suggest that the use of biologic porcine mesh following ELAPER surgery comparatively to myofascial grafts include less operative time, reduced costs related to increased operative time, increased time of hospital stay, plastic surgery involvement, and prolonged side-lying following myofascial skin flap completion (Thomas, 2019). The training involved regarding the use of biologic porcine grafts such as the Cook Biodesign graft used in our case of ACC is minimal as the placement of the graft requires only hydration of the graft and placement without tension as described above.

Our case and the review done by colorectal surgery in a similar area suggests that in vulvar cancer patients biologic graft placement could be considered even if the patient would need radiation in the future (Thomas, 2019). We believe candidates for this procedure would include patients with wide excision making graft closure difficult and patients in which impaired wound healing isn't expected. However, the study done following ELAPER suggests that biologic grafts were equal to that of myofascial flaps even after prolonged chemoradiation to the area which is a known risk for poor wound healing. Further studies are warranted to compare more traditional closure with primary closure and/or surgical flap placement with using biologic grafts in patients with vulvar cancer. Fig. 3.Fig. 4.

Author contributions

KU made significant contributions to the conception, design, and/or acquisition of data, and/or analysis and interpretation of data. MM and JK participated in drafting the article or revising it for important intellectual content and gave final approval of this version to be submitted.



Fig. 3. Vulvar wound during radiation therapy.



Fig. 4. Healed vulva post-radiation.

4. Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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