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Case Report: A Giant Condylomata Acuminata Leading to The Diagnosis of an Underlying Vascular Malformation

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Abstract

Giant condylomata acuminata is a rare intermediate state between benign condyloma and squamous cell carcinoma. They are related to HPV 6 & 11 infections and if left untreated can increase in size and become destructive. This case was managed in a multidisciplinary setting, involving the use of preoperative vascular intervention and surgical resection by plastic surgery and gynaecology teams. Small focuses of early squamous cell carcinoma were present when the surgical specimen was examined. We discuss the important considerations when managing patients presenting with similar chronic lesions and the processes involved in managing this case the way we did.

Introduction

The Giant Condyloma Acuminata (GCA) was first described by Buschke and Löwenstein in 1925 [1]; it is slow growing, locally invasive verrucous plaque; presenting on the penis, and anogenital regions [1]. They're a regional variant of verrucous carcinoma, along with an epithelioma cuniculatum. GCA is associated with HPV 6/11 variants where the E6 protein binds P53 less efficiently when compared to proteins from HPV 16 and 18 [2,3].

If untreated, GCA is locally destructive and infection can spread to pelvic viscera and bony structures. Morbidity rates can be high as recurrence is very common with all treatment modalities [4]. Inadequately treated GCA has a relentless progression and is fatal by involvement of pelvic organs. A complex histological pattern is found containing benign condyloma intermixed with atypical epithelial cells or well differentiated Squamous Cell Carcinoma (SCC). GCA has not previously been reported to be associated with underlying vascular malformations and this is the first reported case within the literature.

Case Discussion

A 38-year old female presented to our plastic surgery service with a fungating lesion to the perineum. Its size had increased over the course of years and it haemorrhaged regularly even in the absence of local trauma. She had unfortunately been in abusive relationships, which prevented her seeking intervention earlier. Venereal diseases tests were negative and the lesion was condylomatous, obscuring her vulva and perineum (Figure 1). An initial biopsy demonstrated some vulval intraepithelial neoplasia but was otherwise consistent with condylomata. A CT scan ruled out metastases, but demonstrated a complex collection of vessels feeding the condylomata (Figure 2).

To further characterise this novel anomaly an MRI with contrast was organised. This scan demonstrated an Arteriovenous Malformation (AVM) with abnormal feeding vessels from the uterine and profunda femoris arteries (Figure 3). Interventional Radiology (IR) assesed the patient for possible percutaneous sclerotherapy but this was abandoned as it was likely to be ineffective and further treatment with arterial embolization was planned.

Super selective angiography using a Progreat micro catheter (Terumo, Japan) followed by embolization using Onyx 34 (Medtronic, US) to the peroneal branches of the right internal iliac arteries via a contralateral approach from left common femoral access was undertaken. A second embolization procedure was undertaken to embolise the supply from the perineal branches of the left internal iliac artery, using an identical technique again from the contralateral side. Combined these embolisations reduced blood flow by approximately 60% (Figure 4). The liquid embolic material Onyx was selected as an embolization agent to occlude the feeding vessels and the small vessels within the lesion to help prevent post procedure large vessel collateralisation.

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Following embolisation the lesion progressed with arterial collateralisation within weeks of interventional procedures. Therefore, after further consult in the MDT the lesion was resected from the perineum and vulva with the use of bipolar, clips and transfixion stitches. The Tantalum powder within the onyx used by the IR team contraindicated monopolar diathermy. The resection plane was to the superficial fascia of the perineum/vulva and anus whilst preserving the vagina, urethra and anus. All the reconstructive options were considered; however, split skin grafting was an adequate reconstruction and permitted monitoring of the surgical site for haemorrhage. Unsurpisingly, this patient had significant bleeding on day 4 and returned to theatre for haemostasis. On day 7 she was discharged, with a graft take of 80%. On further review 6 weeks later, she was fully healed with a pleasing aesthetic result (Figure 5). Her histology confirmed the presence of a condyloma; and foci of SCC.

Discussion

The GCA was first described by Buschke and Löwenstein in 1925 in the penis and they named it "condyloma acuminate carcinoma-like" [5]. It is a rare condition and represents an intermediate state between condyloma acuminata and SCC [6]. They form due the combination of P53 mutation and Human Papillomavirus (HPV) [7]. GCA is more commonly associated with lower risk subtypes of HPV 6 & 11 [8]. It is a slow growing cauliflower like tumour, which is locally aggressive and destructive. It can be differentiated from a standard condyloma, by the presence of "pushing" rather than "infiltrating effect that compresses and displaced the underlying tissues [9]. Both endophytic and exophytic growth patterns are observed, along with undulating papillomatosis of densely keratinised, well differentiated SCC [10].

Various management options are available for these lesions including local laser treatment, Imiquimod, radiotherapy, intralesional interferon alpha. Therapeutic interventions are tainted by relatively low efficacy with a 30-70% recurrence rate identified six months after therapy administration [11]. In large lesions like ours radical excision allows complete histological examination and assessment of tumour free resection margins [12]. Aggressive wide local excision and abdominoperineal resections have been described as options for excisions of this lesions [13].

Abdominoperineal resections are performed rarely in cases where the disease has invaded pelvic organs. Meshed skin grafts are also a recognised reconstructive option of the defect following excision of GCA. CT scans can be used as an investigation looking for distant spread of disease as used in this case [14]. If this patient had extensive disease, we may have changed our surgical management of the GCA. After a comprehensive literature review, we believe this to be the first reported case of a GCA presenting with an associated AVM. A previous report from 2014 described a similarly appearing lesion on a scrotum associated with an AVM [15]. However, this lesion was much smaller than that in our patient and the AVM was not documented with the imaging as in our case.

Result

The patient returned to theatre on day 4 to arrest bleeding points at the site of surgical resection. Skin graft take was 80% on discharge home. This patient was subsequently seen in the outpatient clinic and healed fully maintaining personal hygiene. The specimen contained condyloma along with foci of SCC which was completely excised. She was discussed in the gynaecological MDT and no further intervention was required.

Conclusion

Condylomata is a debilitating illness, which can be overlooked due its benign nature. In severe cases surgical excision should be considered promptly. In longstanding lesions malignancies and associated AVM should be considered and ruled out before definitive management.

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