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Case Report
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A Case of Verrucous Carcinoma of Anus

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Abstract

Background: Verrucous Carcinoma (VC) is a variant of squamous cell carcinoma discovered by Lauren V Ackerman in 1948. It develops relatively slowly, and lymph node metastasis and distant metastasis are rare. It is most commonly found in the oral cavity, vulva, and foot however, there have been reports of VC in the anus. Here we report a case of VC occurring in the anus of a patient who presented with fecal incontinence, which persisted after an Intersphincteric Resection (ISR) for rectal cancer.

Case Presentation: The patient was a 77-year-old man with multiple primary colorectal cancer. He had undergone a laparoscopic right hemicolectomy and intersphincteric resection with a diverting ileostomy. After closure of the ileostomy, he had fecal incontinence and frequent bowel movements in spite of the use of antidiarrhea agents. Anal pain appeared two months after the ileostomy closure. At that point, on physical exam he had multiple warts around the anus, and a biopsy suggested squamous cell carcinoma. Because no distant metastasis or lymph node metastasis was found on computed tomography scan, a skin resection of the perianal region was performed. Postoperatively, he suffered from frequent bowel movements and his fecal incontinence persisted. Multiple warts recurred around the anus three months after the local excision. He underwent an abdominoperineal excision of the rectum. He was discharged on the sixth postoperative day. It has been one year since his surgery and the VC has not recurred.

Conclusion: When a patient presents with a perianal wart and risk factors such as wounds, burn scars, chemical irritation, chronic inflammation, a filthy state due to poor hygiene or incontinence, or HPV infection, it is important to rule out VC. Local excision is considered to be appropriate treatment for VC if there is no obvious metastasis. However, recurrence was observed three months after local excision in this case. Since the anastomosis was closed after an ISR, this recurrence indicates that the margin around the VC lesions may have been insufficient. Thus, it seems that visual examination alone is not thorough enough to assure clear margins and rapid pathological examination of local excision margins is required to assure complete removal of VC cells.

Keywords: Verrucous carcinoma; Anal; HPV

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Abbreviations

VC: Verrucous Carcinoma; SCC: Squamous Cell Carcinoma; HPV: Human Papilloma Virus; ISR: Intersphincteric Resection

Introduction

Verrucous Carcinoma (VC) is a variant of squamous cell carcinoma discovered by Lauren in 1948 [1]. It develops relatively slowly, and lymph node metastasis and distant metastasis are rare [2]. The most frequent sites for VC are the oral cavity, vulva, and foot [3-5], but there are a few reports of VC of the anus. Here we report a case of VC occurring in the anus of a patient with fecal incontinence that persisted after an Intersphincteric Resection (ISR) for rectal cancer.

Case Presentation

The patient was a 77-year-old man with multiple primary colorectal cancer. He had undergone a laparoscopic right hemicolectomy and an intersphincteric resection with a diverting ileostomy. The histopathological examination diagnosed transverse colon cancer (tub2, T3N1 M0 Stage IIIA) and rectal cancer (pap >tub1 >muc, T2N0 M0 Stage I). He was started on a postoperative adjuvant chemotherapy regimen of Capecitabine and Oxaliplatin (CapeOx). After five courses of CapeOx, he developed hand-foot syndrome. The adjuvant chemotherapy was discontinued per the patient's request, and the ileostomy was reversed. Frequent bowel movements and fecal incontinence persisted

Figure 1: Initially, multiple warts were noted around the anus.

Figure 2: At the time of recurrence, a complete excision of the perianal skin including the ISR anastomosis was performed.

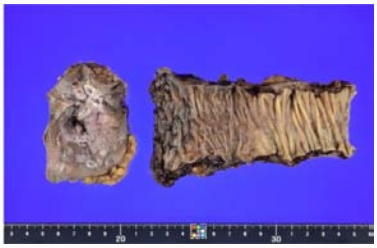

Figure 3: Macroscopic findings at the time of recurrence. 3.1) There was a slightly elevated lesion with a white surface in the anal region.

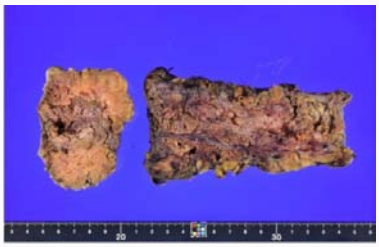

Figure 3.2: There was no tumor exposure in the dissected section.

Figure 3.3: After fixation it was cut out.

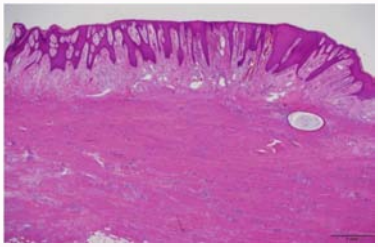

Figure 4: Histopathological findings. 4.1a) Stratified squamous epithelium with thickening, papillary hyperplasia, and extended union of the epithelial processes.

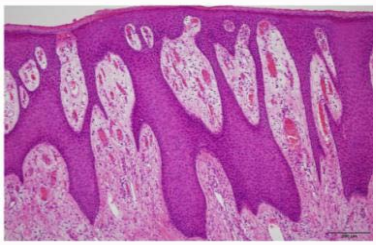

Figure 4.1b: Hyperkeratosis, parakeratosis, and thickening of the granular layer.

Figure 4.2a: CK17 positive.

after the ileostomy closure, and fecal control was not improved despite the use of antidiarrheal agents. He developed anal pain and multiple warts emerged around the anus two months after the ileostomy closure (Figure 1). Biopsy of the warts suggested squamous cell carcinoma. Local excisions of multiple warts were performed with a preoperative diagnose of the limited-disease without metastasis. The histopathological examination showed verrucous carcinoma. Three months after the local excision the warts recurred. On repeat local excision, tumor recurrence was noted on the granulation tissue.

A laparoscopic Abdominoperineal Resection (APR) was performed in two steps. First, the perianal skin was completely excised, including the anastomosis of the ISR, to secure adequate margins of the recurrent tumor (Figure 2). Then the completion APR was performed laparoscopically. The operation time was 266 min, and the amount of blood loss was 45 ml. Macroscopic findings of the resected specimen showed a slightly elevated lesion with a white surface in the anal region without tumor exposure in the dissected section (Figures 3.1 and 3.2). After fixation it was cut out as demonstrated in Figure 3.3. Histopathological findings revealed a stratified thickening of squamous epithelium and papillary hyperplasia, which extended to the union of the epithelial processes (Figure 4.1a). The granular layer was thickened with hyperkeratosis and parakeratosis (Figure 4.1b). Immuno-histologically, the lesion was CK17 positive in the parabasal cell layer and the basal cell layer (Figure 4.2a), and Ki-67 positive in the parabasal cell layer and the basal cell layer (Figure 4.2b). From these findings, we diagnosed it as VC. The presence of koilocytosis around the VC inferred a lesion associated with a condyloma acuminatum (Figure 4.3). The patient was discharged six days after the operation

Figure 4.2b: Ki-67 positive in the parabasal cell layer.

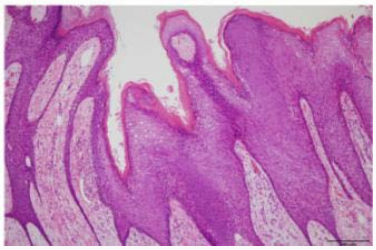

Figure 4.3: The presence of koilocytosis around the VC.

without postoperative complications. One year after the operation, no recurrence has been observed.

Discussion

Verrucous Carcinoma (VC) is a variant of squamous cell carcinoma with distinctive clinical and histologic features. Typically VC appears with an irregular and cauliflower-like appearance, but not in this case. Histopathologically, VC is characterized by prominent keratinization, is highly differentiated, and has a papillary and scaly shape. It tends to proliferate outward. Immuno-histologically, VC is characterized by a positive basal cell layer on Ki-67 staining [6].

Risks for the onset of VC include injury, burn, chemical stimulation, chronic inflammation, poor hygiene or incontinence, and HPV infection [7]. In this case, the patient developed a filthy state in the perianal region due to fecal incontinence which led to chronic inflammation and may have triggered VC. Zidar et al. [4] reported that anal VC is not associated with HPV infections and must be distinguished from Buschke-Lowenstein tumor, which is associated with low-risk HPV. However, De Vuyst et al. [8] reported that 74.9% of men with anal carcinoma have HPV. Further, histopathological examination of our patient's lesions revealed koilocytosis, which is pathognomonic for low grade squamous intraepithelial lesions and thus HPV infections. HPV infections have been reported in other cases of VC. Therefore, we think that further accumulation and classification of VC cases is necessary to clarify the relationship between anal VC and HPV infection.

For the treatment of VC, surgical resection of the tumor is preferred, and radiation therapy should be avoided due to the risk of malignant transformation [2,9,10]. A good prognosis can be expected after treatment a local excision because the development of VC is very gradual and the occurrence of metastasis is rare. In the present case, we experienced a local recurrence shortly after the first excision. This indicates it is important to secure negative surgical margins during the tumor excision by adding intraoperative rapid pathological diagnosis; insuring full eradication of the VC and decreasing the risk of recurrence.

Conclusion

We reported a case of VC occurring in the anus of a patient who presented with fecal incontinence that persisted after an Intersphincteric Resection (ISR) for rectal cancer. In this case, chronic fecal incontinence created a filthy state and chronic inflammation. It is crucial to keep in mind the possibility of occurrence of VC around the anus. During surgical resection, it is necessary to confirm that the margins are cleared by local excision in order to decrease the risk of VC recurrence.

Authors' Contribution

HK and YH wrote the manuscript. SY supervised the case. All other authors reviewed the manuscript. All authors read and approved the final manuscript.

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