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Case report

Case report of an anal adenocarcinoma arising from a perineal lump

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HIGHLIGHTS

- Although anal adenocarcinoma is rare, it can arise from chronic inflammatory states such as long standing perianal fistulae.
- MRI can be helpful in delineating the underlying aetiology, but histology is ultimately required for definitive diagnosis.
- Radical surgery with either neoadjuvant or adjuvant chemoradiotherapy appears to be most effective.

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ABSTRACT

Anal adenocarcinoma is a rare condition and can arise in chronic inflammatory states such as in Crohn's disease, or in a chronic fistula-in-ano. We report our diagnosis and management of a patient who presented with a large perineal lump with a long-standing history of perianal fistulous disease. This was initially evaluated with a Magnetic Resonance Imaging, and the diagnosis was confirmed with biopsy. Multimodality treatment with chemoradiotherapy and surgery should be offered to achieve the best outcomes.

1. Introduction

Anal carcinoma is a rare condition, accounting for only 1% of cancers of the gastrointestinal tract [1]. Amongst anal cancers, a large majority are histologically squamous cell carcinomas, and only about 10% of cancers in this region are adenocarcinomas [2]. These are thought to originate from the columnar epithelium lining the anal glands [3]. As defined by WHO criteria, anal adenocarcinomas arise from three sites, the rectum, anal glands, and a chronic fistula-in-ano [4]. Adenocarcinomas arising from a chronic fistula-in-ano are rare, and constitute only about 7% of anal canal carcinomas [5].

The diagnosis of cancer in patients presenting with chronic anal fistula requires a high index of suspicion. Together with imaging and photographs, we describe in this case report our method of diagnosis and management of a patient with anal adenocarcinoma on a background of a chronic anal fistula. We also discuss existing evidence in the management of this rare disease.

2. Case report

We report a 72 year old gentleman who presented with a 5 year history of an enlarging lump at the perineal region (Fig. 1). The lump started as a small red papulae but gradually grew in size. He had never sought prior medical attention for this lesion, and only did so at the current instance as he was experiencing increasing discomfort. He was not known to have any perianal disease. There was also no family history of malignancy. On examination, the lump was non-tender, measured about 3 cm in size, and was firm in consistency. Digital rectal examination did not reveal any mass in the anal canal or distal rectum.

To evaluate this mass better, a Magnetic Resonance Imaging (MRI) of the pelvis was performed. The lesion measured 2.7 × 4.2 × 2.6 cm and appeared hyperintense on T2−weighted imaging (Fig. 2). There were also several other transphincteric and intersphincteric fistula tracts identified (Fig. 3). Incidentally, a large polyp was also noted at the distal sigmoid colon. Small volume inguinal and pelvic lymph nodes were also visualized.

A colonoscopy was performed subsequently. The aforementioned sigmoid polyp was identified and biopsied Fig. 4). Colonoscopy was otherwise unremarkable with rectal mucosa and the

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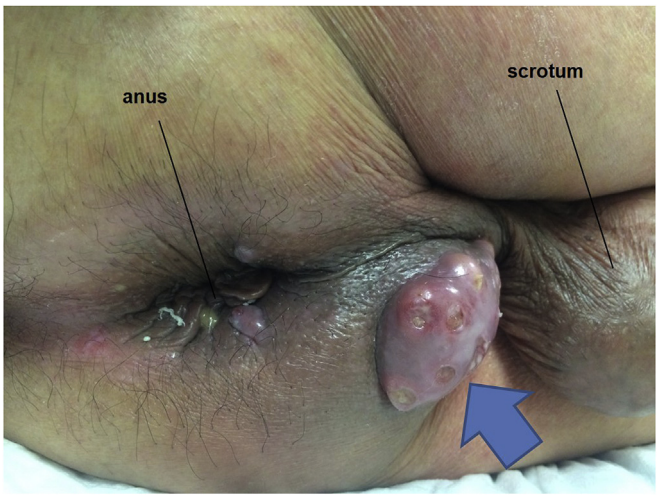

Fig. 1. Perineal lump with pits on the surface draining hemo-purulent fluid (Arrow).

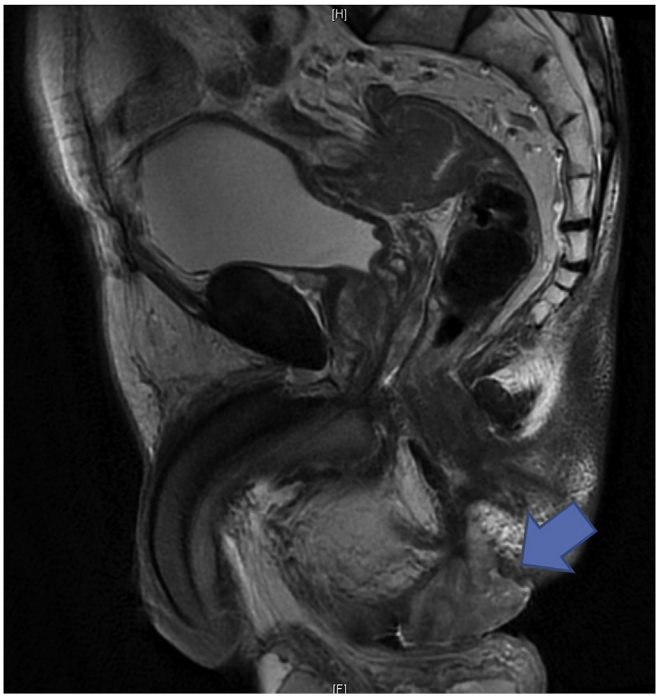

Fig. 2. Perineal lump as visualised on MRI (Arrow).

anal canal appearing normal. Multiple biopsies of the lump were taken externally, and small amounts of haemo-purulent fluid was expressed during the biopsy.

The biopsy of the perineal mass actually demonstrated fragmented portions of squamous mucosa and stroma with glandular neoplasm, ulceration, necrosis and granulation tissue. The neoplasm was composed of infiltrative glandular structures with colonic-type features (CDX2 and CK20 positive, CK7 negative), leading to a diagnosis of adenocarcinoma with colorectal origin. Biopsy of the sigmoid colon polyp was consistent with a tubulovillous adenoma of low grade dysplasia. This case was discussed at a multidisciplinary tumour board and neoadjuvant chemoradiation therapy prior to definitive abdomino-perineal resection was advised. He was planned to receive capecitabine, as well as 25 fractions of 32 Gy radiotherapy.

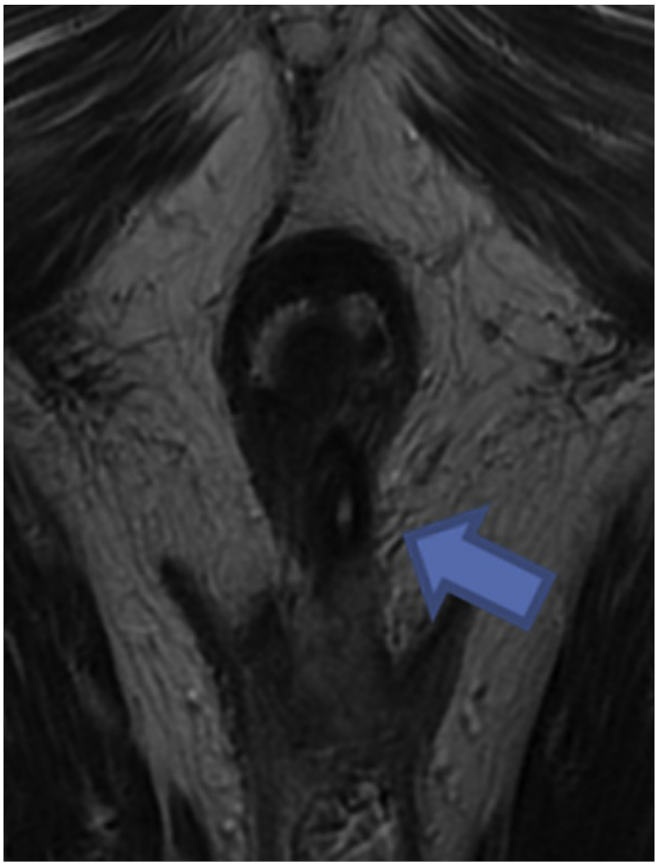

Fig. 3. Sequential coronal sections on MRI showing a fistulous tract (Arrows).

3. Discussion

The pathogenesis of a neoplastic lesion from the anal glands is due to secondary change following chronic inflammation resulting in persistent mucosal regeneration [6]. This has been implicated following the observation of anal adenocarcinoma arising in patients with chronic local inflammatory states, such as anal fistulas, as well as in inflammatory bowel disease (IBD). Other postulations for the origin of anal adenocarcinoma include the implantation of cancer cells from a more proximal location along the colon [7], or the use of chronic immunosuppression in patients with IBD [8]. A meticulous evaluation of the gastrointestinal tract during colonoscopy is therefore important, and all polyps should be biopsied for histological evaluation. The diagnosis of anal adenocarcinoma requires a high index of suspicion in order to avoid delay in treatment. The absence of any antecedent gastrointestinal tract cancers, as well as a long history of symptoms of the fistula, strongly suggest the presence of cancer arising de novo from a previously benign anal fistula [9]. The diagnosis can however only be confirmed on histological biopsy as the differentials include conditions such as tuberculosis, actinomycosis, perianal Paget's disease, as well as lymphogranuloma venereum [10].

A lack of consensus exists regarding the most optimal modality of treatment for patients with this condition. Some possible approaches include primary surgery with adjuvant chemoradiotherapy (CRT), primary CRT, as well as neoadjuvant CRT with surgery and adjuvant CRT [11]. The largest published series of 82 patients compared 5-year local recurrence rates and 5-year disease free survival in 45 patients with radiotherapy and surgery, primary

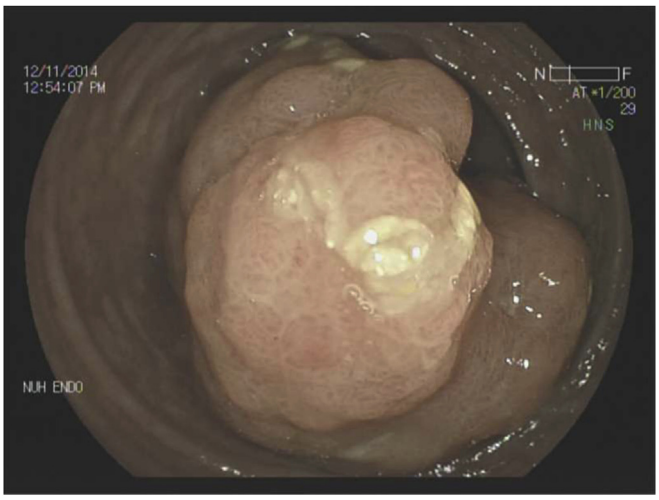

Fig. 4. Sigmoid polyp.

CRT, and surgery alone. Recurrence rates were 37, 36, and 20% respectively, and disease free survival rates were 25, 54 and 22% respectively [12].

Other studies have however shown that multimodality therapy including abdomino-perineal resection (APR) with CRT appear to provide the best results. One study at the MD Anderson Cancer Centre showed that primary CRT was associated with high rates of local recurrence and distant metastases, and should be combined with APR [13]. Another study compared the 5-year survival rates of patients treated with APR alone, CRT alone, APR with CRT, and no treatment. Results were 34, 0, 38, and 0% respectively [14]. Contemporary studies have suggested that chemoradiotherapy should be administered before abdominoperineal resection in order to achieve R0 resections and negative margins. Hongo et al. showed 85% (6/7) complete response rates after neoadjuvant therapy [5]. These findings were further supported in another study whereby positive circumferential resection margins during abdominoperineal resection was present in only 8% (1/13) of patients following neoadjuvant therapy [15].

4. Conclusion

We report a patient with a chronic, progressive perineal lump which was confirmed to be anal adenocarcinoma on the background of a chronic anal fistula on histology. Current literature suggests that when the disease is potentially curative, radical surgery with either pre- or post-surgery chemoradiotherapy should be attempted to achieve the best overall survival.

Ethical approval

No institutional review required as per guidelines as this is a case report.

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All authors have obtained no support from any organisation for the submitted work; no financial relationships with any organisations that might have an interest in the submitted work, and no other relationships or activities that could appear to have influenced the submitted work. Verbal consent was obtained from the patient for this case report.

Conflicts of interest

The authors disclose no conflicts.

Guarantor

The corresponding author Dr Tan Ker Kan is the guarantor.

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