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BCG mycotic aneurysm: Case report on a rare entity

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A B S T R A C T

Mycotic aortic aneurysms (MAA) are rare, accounting for 0.6–2.0% of all aortic aneuryms. MAA secondary to intravesical BCG instillations are even rarer, with less than a hundred reported cases till date. Given the delayed presentation, non specific presenting symptoms and significant risk of mortality (90% without intervention, 10.3–22.7% with intervention), diagnosing this complication is challenging.

1. Introduction

Bacillus Calmette-Guerin (BCG) is a live attenuated strain of *Mycobacterium bovis* that was developed initially as a vaccine against tuberculous mycobacteria infections. Since then, many studies have proven the efficacy of repeated intravesical instillations of BCG in reducing recurrence of non-muscle invasive urothelial carcinoma of the bladder, particularly in high risk categories (high grade cancer, T1 cancer, carcinoma in situ).[1] Shelley et al. has shown that maintenance BCG reduce risk of tumour recurrence (OR 0.61–0.68) as well as progression (OR 0.73).[1]

However, intravesical instillations of BCG is not without its complications. Side effect rates are high, ranging from mild, localised inflammatory responses to severe systemic complications that may be fatal. Table 1 lists some of the more commonly known adverse effects associated with intravesical BCG use as well as its management.

Mycotic aortic aneurysms (MAA) are rare, accounting for 0.6–2.0% of all aortic aneuryms.[2] MAA secondary to intravesical BCG instillations are even rarer, with less than a hundred reported cases till date. Given the delayed presentation, non-specific presenting symptoms and significant risk of mortality (90% without intervention, 10.3–22.7% with intervention), diagnosing this complication is challenging.[3]

2. Case study

A 76-year-old man was diagnosed with TaHG bladder cancer in November 2018 and was prescribed a weekly instillation of 6-week induction intravesical BCG therapy as per standard protocol, completed in May 2019.12.5mg oncoTICE BCG containing 2–8x10^8 colony forming units of Tice BCG was instilled throughout treatment. Post induction bladder biopsy was negative, after which this patient went onto monthly maintenance intravesical BCG instillations.

However, after 4 doses of maintenance BCG he complained of shortness of breath, increased work of breathing and palpitations, with associated nausea, vomiting and fevers. His condition deteriorated rapidly and he went into type I respiratory failure with increasing oxygen requirements. Fig. 1A is a CXR demonstrating widespread bilateral patchy lung consolidation. Respiratory was consulted, and he was admitted into intensive care unit (ICU) and intubated. His condition gradually improved over a 14-day admission period. Diagnosis given at the time was acute respiratory distress syndrome (ARDS) secondary to severe allergic pneumonitis from BCG. Maintenance intravesical BCG therapy was ceased indefinitely. Infectious diseases (ID) was consulted during the admission and urine and blood cultures taken came back positive for M. bovis. However, in light of the patient's significant recovery without antibiotics he was not initiated on full spectrum antituberculosis therapy.

This patient went on to have a left middle cerebral artery territory infarct in April 2020 as well as a right percutaneous nephrolithotomy in August 2020, recovering well from both. Two computerised tomography (CT) scans of his abdomen performed during this period did not demonstrate any abnormalities. In October 2020, he re-presented to the Emergency Department with low back pain, nausea, vomiting, weight loss of more than 15kg over the preceding two months. Bloods performed were grossly unremarkable with WCC 5.48, CRP 25, ESR 16 with a slight lymphopaenia of 0.39. Repeat CT imaging revealed a new 45mm enhancing mass sandwiched between the descending abdominal aorta and the inferior vena cava (IVC) with an associated infrarenal aortic aneurysm. A separate 24mm mass was seen along the right common iliac vessels (Fig. 1B). CT guided biopsy confirmed M. bovis consistent with

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Table 1
Side effects of intravesical BCG instillation and management.[5]

Unwanted Adverse Effects (frequency)	Management*
Local (62.8%)	
Irritative lower urinary tract symptoms (23.6–35%) - urinary urgency, frequency, dysuria	• analgesia • urine culture + antibiotics • cystectomy if chronic and severe
Haematuria (22.6%)	• urine culture + antibiotics • cystoscopy if persistent
Symptomatic granulomatous prostatitis	• urine culture + antibiotics
Epididymo-orchitis	• urine culture + antibiotics • orchidectomy if chronic
General (30.6%)	
Malaise, fever (8.1–15.5%)	• conservative • anti-pyretics
Arthralgia/arthritis	• NSAIDs • corticosteroids
BCG sepsis (0.3%)	• cessation • CXR, blood culture, urine culture • anti-tuberculosis therapy • corticosteroids if persistent
Allergic reaction	• antihistamines • NSAIDs/corticosteroids

*Management includes cessation of therapy and consideration of anti-tuberculosis therapy for all adverse effects that are progressive or persistent.

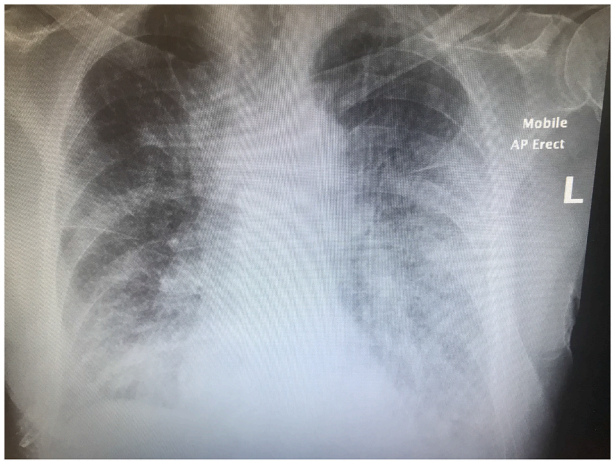

Fig. 1A. Severe allergic pneumonitis secondary to intravesical BCG.

BCG. Given the patient's comorbidities, decision was made to perform an endovascular aortic repair (EVAR) rather than open surgical resection and debridement. There were no immediate postoperative complications. He required a period of ICU admission for inotropic support and close monitoring. He was also put on a long term schedule of isoniazid, rifampicin and ethambutol treatment for BCGosis. Repeat CT imaging over time indicated slow resolution of the offending mass (Fig. 2). Patient declined any further intravesical therapy and opted for imaging and cystoscopy surveillance.

3. Discussion

Intravesical instillation of BCG post transurethral resection of bladder tumour (TURBT) has been the gold standard therapy for non-muscle invasive high risk bladder cancers. This reduction in tumour progression and recurrence stems from local T-cell mediated immune

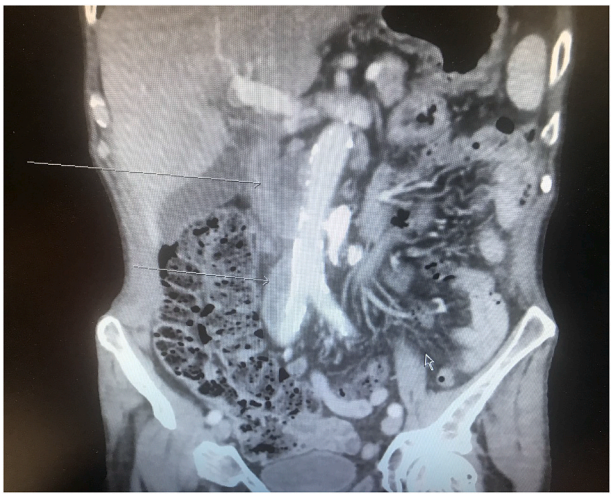

Fig. 1B. CT demonstrating abovementioned two new mycotic lesion between aorta and IVC as well as along right iliac artery respectively.

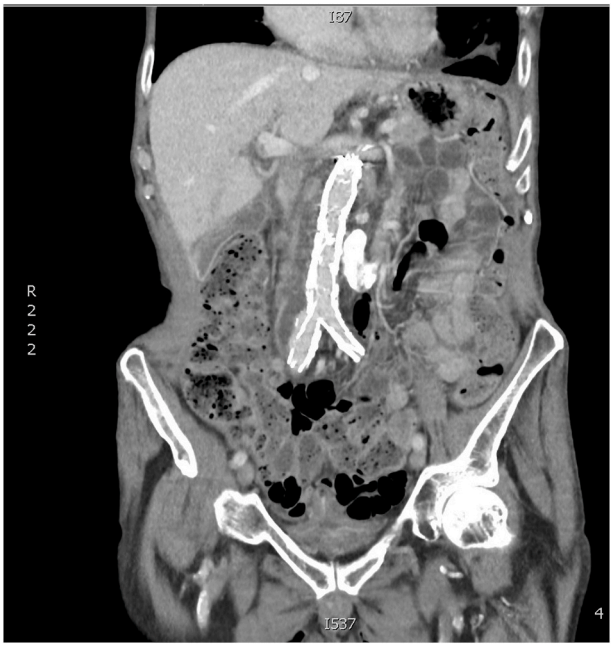

Fig. 2. CT demonstrating resolution of mycotic aneurysm post endovascular repair.

inflammatory response and tumour necrosis factor (TNF) release.[1] Its mechanism is not completely understood.

The mechanism behind development of a MAA from BCG intravesical instillation is likely one of BCGosis, contiguous spread through the vasa vasorum, leading to thinning of the tunica intima and media and resulting aneurysm.[4] Treatment is mandatory to reduce risk of aortic leak/rupture. Surgical resection of the offending lesion with placement of an interposition graft or a bypass with long term triple antibiotic therapy has been the gold standard treatment for such cases.[4] However, EVAR has become increasingly popular given its lower overall morbidity and mortality rate perioperatively. Studies comparing between the two treatment options are scarce and are mainly single center, retrospective, with small study populations.[4] Regardless, prognosis is poor with diagnosis of MAA. There is some evidence to show that EVAR offers superior short term survival as compared to open surgical resection (OSR).[5] Infection related complications are more frequent with EVAR, with a recent multicenter study of 120 MAA managed with EVAR

demonstrating 5% risk of subsequent OSR, 27% risk of infection related complications, and a 41% survival rate over a 5-year follow up period.[4]

Pre-disposing factors postulated include underlying anatomical abnormalities such as aortic coarctation, valvular deformities or presence of prosthetic arterial devices in situ.[4] Existing atherosclerotic plaques and aneurysm/pseudoaneurysm also increases the risk of developing MAA with treatment.[5] Contraindications of intravesical BCG therapy should also be adhered to so as to reduce risk of BCGosis and MAA.

4. Conclusion

In summary, we report a recent incident of a patient with MAA secondary to recent intravesical BCG treatment for bladder urothelial cell carcinoma. The incidence of such cases is low, and may represent significant under reporting given the non-specific nature of its presentation with potentially disastrous consequences. Screening for pre-existing cardiovascular comorbidities and appropriate counselling should be considered for high risk patients prior to commencing BCG therapy.

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