Perineal xanthogranulomatous pseudotumor due to intermittent catheterization: a liposarcoma mimic. Case report

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A large perineal fatty tissue mass shown to be an encapsulated xanthogranulomatous reaction was found in a spinal cord injured man whose neuropathic bladder was for a long time being managed by clean intermittent catheterization, but who had chronic urinary tract infection. He was also known to be a chronic alcoholic. We would say that an alcohol numbed sensorium and multiple catheterizations led to recurrent small urethral perforations which in the setting of chronic bacteriuria promoted the unusual inflammatory mass. Appreciating the increasing use of clean intermittent catheterization for the spinal cord injured, we suspect that similar cases of 'xanthogranulomatous pseudotumors' will be found, and therefore we describe this most unusual clinical presentation.

Keywords: perineal xanthogranuloma; catheterization; complication.

Introduction

The perineum is a unique soft tissue region which lends itself to ease of physical examination. Although primary malignancies of the perineum are rare, any mass found in this region will lead to considerable anxiety. We briefly review perineal masses and add to this list a most unusual mass which we believe to be the first reported case of a perineal xanthogranulomatous pseudotumor.

Case report

A 62 year old male SCI patient had bladder function maintained by clean intermittent self catheterization (ISC) for the past 7 years. He had a daily consumption of 6–8 liters of wine, but was in good general health when he presented with a painless perineal mass. This progressively enlarging mass of 4–6 weeks duration was unassociated with fever, weight loss, malaise or hematuria. On examination, a firm midline perineal mass of approximately 3 × 5 cm was found and it was fixed, noncompressible, and nontender. Subsequent retrograde urethrography, intravenous pyelography, and computed tomography were unremarkable other than revealing a small scarred right kid-

ney, compatible with chronic pyelonephritis. Laboratory studies including CBC, electrolytes, creatinine, and BUN were within normal limits. There was a slight abnormality of the liver function tests. Persistent silent chronic bacteriuria (> 100 K *E. Coli* and > 100 K *S. viridans*) was again found.

After detoxification and lengthy counselling, surgical exploration was recommended, with a preoperative diagnosis of possible liposarcoma. The perineum was opened through the midline, and a well-encapsulated fatty mass surrounding the bulbar urethra was uneventfully excised (Fig 1).

Histologically the tissue showed a predominance of lipid laden macrophages (foam cells)

Figure 1 Fibro-fatty perineal mass.

Figure 2 (a) Low power view of perineal mass. Note the fat and inflammatory cells. (b) High power magnification showing foam cells, lymphocytes, giant cells, plasma cells, and fat.

along with fibroblasts, lymphocytes, plasma cells, giant cells, and fat consistent with the diagnosis of xanthogranuloma (Fig 2). On follow up examination 6 months later, there was no evidence of recurrence. He had remained alcohol free during this interval.

Discussion

It is rare to find a nontender mass in the perineum and therefore finding one generated considerable anxiety.

Although there are many reports of perineal masses, most have been sporadic case reports. Neoplasms of the extraperitoneal rectum and anus may present as a palpable mass in the perineum, but are rare. Mangiante *et al* reported a series of 120 patients with biopsy-proven neoplasms of anorectal origin, but none presented as a perineal mass.¹ Perianal or anal margin carcinomas arising outside the anal verge have the same potential pathology as other skin malignancies, such as squamous cell carcinoma and melanoma. Their initial presentation as a perineal mass is also rare.² Gritsch *et al* reported the case of a giant condylomata acuminata of the perineum, but such occurrence is unusual and this diagnosis is easily made by the characteristic exophytic appearance.³ A number of cystic structures may present as a perineal mass, including ectopic ureterocele,⁴ dilatation of Cowper's gland duct,⁵ urethroperineal cyst from a posterior urethral duplication,⁶ and a clo-

acogenic cyst.⁷ These are also case reports, and are of rare occurrence. Liposarcomas, being the most common of the soft tissue sarcomas, have also been reported in the perineum.⁸ Steeper *et al* reported a series of nine patients with angiomyxoma, two of whom presented as a perineal mass.⁹ Other reported rare entities include periurethral neurofibroma,¹⁰ granular cell tumor¹¹ and perineal infiltration by plasma cells in an individual with multiple myeloma.¹²

We were unable to find any previous report of a perineal mass secondary to a xanthogranulomatous reaction. Xanthogranulomatous pyelonephritis (XGP) is a well established pathological entity, but search of the literature fails to show any other similar case. XGP is a chronic form of pyelonephritis, usually associated with calculi or obstruction, and often with a clinical presentation suggestive of a renal mass. The typical histological appearance is characterized by a cellular infiltrate of lipid laden mononuclear macrophages (foam cells) in the diffuse background of adipose and fibrous tissue very similar to the histological findings seen in our patient.

The exact etiology of XGP is unknown. Venous occlusion, hemorrhage, lymphatic obstruction, prolonged antibiotic therapy with chronic renal obstruction and infection, altered immune status, impaired cellular immune response, abnormal lipid metabolism, renal ischemia, diabetes and malignancy have all been suggested as contributing to a xanthogranulomatous inflammatory

response. Organisms frequently implicated in XGP are *E. coli* and *P. mirabilis*, and the calculi are usually composed of struvite.¹³

In this report, we suggest that the xantho-granulomatous inflammatory reaction was the result of recurrent trauma to the urethra from chronic ISC in the setting of numbed sensorium and a state of high urine output requiring multiple catheterizations. The

presence of chronic bacteriuria was also likely to be a contributing factor. Since ISC is being used increasingly in SCI patients, we suggest that similar cases will probably be identified. Hopefully, with a preoperative diagnosis of possible perineal xantho-granulomatous pseudotumor the serious anxiety associated with a potential liposarcoma will be alleviated.

References

- 1 Mangiante EC, Dilawari RA, Britt LG (1983) Neoplasia of the extraperitoneal rectum and anus. *Am Surg* **49**: 73–75.
- 2 Waugh DE (1986) Anal and perianal malignancies. *Surg Clin North Am* **66**: 841–844.
- 3 Gritsch HA, Randazzo RF, Layfield LJ, DeKernion JB (1989) Invasive giant condylomata acuminata: A case report. *J Urol* **141**: 950–952.
- 4 Rocereto TF, Campbell WA III (1980) Ureterocele presenting as a perineal cyst. *Obstet Gynecol* **55**: 54–56.
- 5 Redman JF, Rountree GA (1988). Pronounced dilatation of Cowper's gland duct manifest as a perineal mass: A recommendation for management. *J Urol* **139**: 87–88.
- 6 Anderson RL, Altman DH, Lankau CA, Lopez P (1980) Urethroperineal cyst in a newborn: Posterior urethral duplication. *AJR* **135**: 622–624.
- 7 Anani P, Leu F, Delacrétaz J (1983) Perineal cloacogenic cyst. *Dermatologica* **166**: 104–106.
- 8 Hulbert JC, Rodriguez PN, Cummings KB (1984) Perineal liposarcoma: Diagnosis and management. *J Urol* **131**: 1185–1187.
- 9 Steeper TA, Rosai J (1983) Aggressive angiomyxoma of the female pelvis and perineum. *Am J Surg Pathol* **7**: 463–475.
- 10 Selli C, Villari D, Amorosi A et al (1987) Periurethral neurofibroma. *Urology* **30**: 82–83.
- 11 Hansborg N, Refn H (1981) Granular cell tumor in the perineum. *Acta Obstet Gynecol Scand* **60**: 219–221.
- 12 Lewinski UH, Bayer I, Turani H (1984) Perineal infiltration by plasma cells in a patient with multiple myeloma. *Dis Colon Rectum* **27**: 479–480.
- 13 Petronic V, Buturovic J, Isvaneski M (1989) Xanthogranulomatous pyelonephritis. *Br J Urol* **64**: 336–338.