Inter-Osseous Epidermoid Inclusion Cyst Of The Sacrum: A Case Report

Mike Parry[1], Anish Patel[2], Rajesh Botchu[2], Vaiyapuri S[3], Desai[4], Jeys Lee[1]

Abstract

Whilst common in subcutaneous locations, inclusion epidermoid cysts arising from the pelvic bones are extremely rare. We report the case of a patient presenting with coccydynia and altered bowel habit who, following investigation was found to have a large cystic lesion of the sacrum with extending anteriorly into the presacral tissue resulting in compression of the rectum. The lesion was treated by surgical resection, sacrificing the lower sacral nerve roots. Histology confirmed an epidermoid cyst. Whilst a rare presenting symptom, careful assessment of the patient presenting with coccydynia and altered bowel function must raise the suspicion of a sacral pathology that demands further investigation.

Keywords: sacral epidermoid cyst, rare presentation

Introduction

Epidermoid cysts are common benign lesions that most commonly occur in subcutaneous locations. The commonest age at presentation is between 19 and 45 years although they can occur in children in whom 50% of cases are associated with Gardner's syndrome. Common sites include the trunk, neck, face, hands and feet, whilst in the head and neck, it is the most common subcutaneous cyst (1). Theories as to the etiology of epidermoid cysts depend on trauma, resulting in implantation of epithelial elements into the subcutaneous structures, or due to a congenital cause due to the inclusion of epidermoid elements during tube closure during embryogenesis. The rarity of these lesions following common orthopaedic procedures would suggest against the traumatic or iatrogenic cause of inclusion. Epidermoid inclusion cysts in the pelvis, sacrum and coccyx are, however, extremely rare with only a few previously reported cases (2). We report the case of a large epidermoid inclusion cyst of the sacrum with anterior expansion to involve the pararectal fascia, rectum and sacral nerve roots.

Case Report

39 year old male, presented with an 11 month history of coccydynia and weight loss, insidious in onset and without any antecedent history of trauma. The pain was aggravated by sitting and weight bearing. At presentation, he was noted to be constipated though did not describe any alteration in his urinary function. He noted parasethesiae when he crossed his legs and an aching discomfort throughout both legs after walking a short distance. Examination revealed tenderness to the left of the midline in the natal cleft. Digital rectal examination deciphered a large tender, extraluminal retrorectal swelling. Investigations comprised of plain radiography, magnetic resonance imaging (MRI) and contrast enhanced computerized tomography (CT). MRI demonstrated a lobulated 8 x 10 x 13 cm predominantly cystic lesion involving the sacrum starting from the level of the third sacral segment and extended caudally to involve the other sacral segments with a large component extending anteriorly into the presacral tissues. This caused compression of the rectum and consequent proximal large bowel obstruction. No fistula was demonstrated with contrast enhanced CT. No connection of the mass lesion to the dermis or subdermis was identified. On MRI the lesion was isointense to skeletal muscle on T1 weighted images and predominantly hyperintense on T2 weighted images with areas of heterogeneity. Initial haematological investigations, including erythrocyte sedimentation rate and C-reactive protein, were non-contributory. Tissue diagnosis was by CT guided biopsy of the sacral mass with the patient in the prone position using a midline approach. Histopathological examination confirmed features consistent with an epidermoid cyst without evidence of malignancy. Following the biopsy, the patient developed an infection of the biopsy tract and persistent discharge from the cyst cavity. Subsequent to undergoing biopsy of the sacral lesion, the patient developed urinary retention due to compromise of the second and third sacral nerve roots bilaterally. The patient elected to undergo surgical resection to remove the cyst. This was performed using computer-navigation

[1]Department of Orthopedic Oncology. [2]Department of Musculoskeletal Radiology, [3]Department of pathology The Royal Orthopedic Hospital, Bristol Road South, Northfield, Birmingham, UK
[4]Department of Surgery, The Queen Elizabeth Hospital, Birmingham, UK

Address of Correspondence
Dr Rajesh Botchu
Department of Musculoskeletal Radiology,
The Royal Orthopedic Hospital
Bristol Road South
Northfield, Birmingham, UK
Ph-00447967505057
Email – drbrajesh@yahoo.com


Dr. Anish Patel


Dr. Rajesh Botchu


Dr. Mike Parry


Dr. Jeys Lee

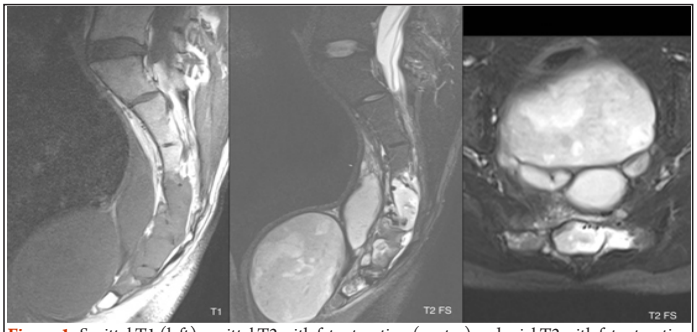

Figure 1: Sagittal T1 (left), sagittal T2 with fat saturation (center) and axial T2 with fat saturation (right). This shows a lobulated mass centered over the sacrum originating at the level of S3 and extending both caudally and anteriorly. The lesion is isointense to skeletal muscle on T1 weighted imaging and is heterogeneously hyperintense on T2 weighted sequences which gives a slightly speckled appearance. The large anterior component of the lesion compresses the rectum.

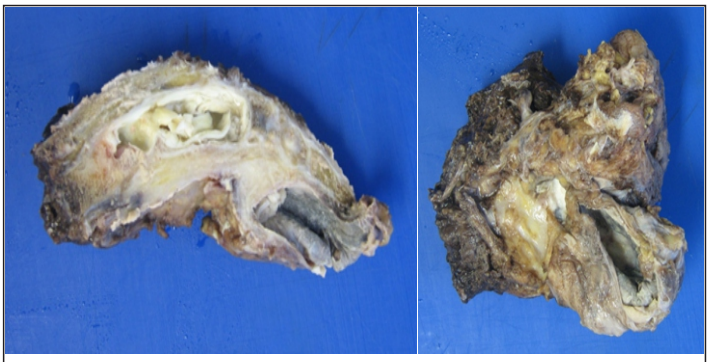

Figure 2: Gross specimen following sacrectomy demonstrating a well defined cystic lesion within the sacrum containing whitish caseous material. The lesion expands anteriorly and has been resected with a section of rectum.

assisted surgery through a posterior midline approach. A meticulous dissection was performed to resect the sacrum and coccyx with an osteotomy performed through the second sacral vertebra and sacrificing the lower sacral nerve roots. Through an anterior approach, the cyst was mobilized together with the resected sacrum and a rectal resection was performed. A defunctioning colostomy was fashioned through the anterior abdominal wall and a suprapubic urinary catheter inserted. No plastic surgical coverage was required to fill the posterior parasacral defect. Histopathological examination of the resection specimen demonstrated. The patient made an uneventful recovery and is currently pain free and mobile. He remains with a suprapubic urinary catheter and a colostomy.

Discussion

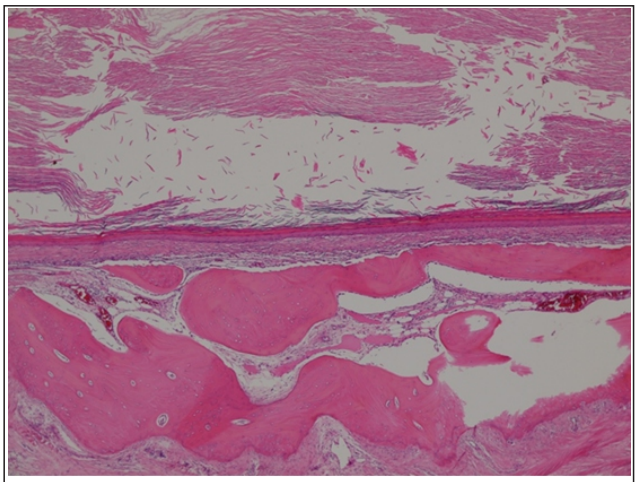

Figure 3: Histology specimen demonstrating an intraosseous squamous lined cyst containing keratinous debris. There is evidence of chronic inflammation in the cyst wall but no evidence of malignancy. (Haematoxylin & Eosin x100).

Epidermal cysts can be described as a dermal cystic enclosure of keratinizing squamous epithelium that is filled with keratin. Theories as to the cause of epidermoid cysts either rely on trauma or surgical insult resulting in implantation of epithelial tissue into the subcutaneous tissues (3,4). An alternative theory relies on abnormalities in embryogenesis with misplacement of epithelial tissues during neural tube closure (2,5). Finally, an alternative theory is one of penetration of an existing mass into deeper structures (6). In this case, the patient presented without a cutaneous mass or cyst but rather developed a deeper mass with expansion anteriorly. There was no history of antecedent trauma or inoculation, which would suggest a congenital aetiology for development of the cyst. Epidermoid cysts can be difficult to characterize on MRI as the imaging features can be variable (7). The lack of surrounding tissue oedema, in the absence of concurrent infection, can differentiate epidermoid cysts from osseous malignancy. They demonstrate well-defined limits and peripheral enhancement on post contrast images. However, the variability in signal intensity, which may be the result of variations in cholesterol content or the ratio of keratin to cholesterol within the cyst, can make the diagnosis on MRI difficult. The typical presentation of these cysts are of a painless, slow growing, well-circumscribed swelling (8). However, cysts in this area are extremely rare, and presentation with obstructive pelvic symptoms has not been recorded previously. The expansion of the cyst to involve adjacent anterior intrapelvic structures highlights the erosive nature of these cysts and may demonstrate the presence of inflammation or infection within the cyst, as manifest by the involvement of the sacral nerve roots. The presentation of the cyst in this case is not typical. Previous reports of intraosseous epidermoid cysts have not demonstrated neurological involvement or obstructive symptoms (2), but rather patients with epidermoid cysts in the retrosacral and paracoccygeal regions present with pain and coccydynia (9). Whilst pain was certainly a feature in this presentation, the presence of an alteration in bowel function prompted presentation, rather than coccydynia. The differential diagnosis at presentation was of a primary osseous malignancy, most likely a sacral chordoma, or a Tarlov cyst. However, these differentials were refuted by initial biopsy and the final histology confirmed on the resection specimen. Whilst epidermoid inclusion cysts are not uncommon in the extremities, head and neck and trunk, an intraosseous presentation within the sacrum is extremely rare. The involvement of sacral nerve roots and inflammatory erosion anteriorly, as seen in this case, to our knowledge has not been previously reported. The significant morbidity associated with resection of this benign lesion, the requirement for a defunctioning colostomy and a permanent supra-pubic catheters as well as the obliteration of erectile function highlights the potential for aggressive local infiltration of these lesion

when present in an intraosseous location. Careful assessment of patients presenting with pain and obstructive large bowel symptoms must be mandatory as digital rectal examination in this case, at an early stage and subsequent urgent cross sectional imaging may have identified the lesion at a stage where surgical resection without such morbidity may have been possible.

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