Intractable coccygodynia is frequently labelled as being idiopathic or post-traumatic in the literature (1, 2). Herein, we report a case of epidermal inclusion cyst in the preoccygeal region causing severe pelvic and coccygeal pain.

Case report

A 33-year-old pregnant woman was admitted to our hospital complaining of severe pelvic and coccygeal pain. Her medical examination and laboratory tests were found within normal limits. In order to explain her pain, initially a pelvic ultrasound was performed which revealed a huge hypoechoic cystic mass in the preoccygeal-presacral region. She then underwent a pelvic magnetic resonance imaging (MRI) examination in order to better delineate the characteristics and extension of this huge mass. On these images the mass was hypointense on T1 weighted images and extremely hyperintense on T2 weighted images. We also performed a diffusion weighted sequence which exhibited high signal intensity for the mass. We thought that this finding could be suggestive of an epidermal inclusion cyst similar to that of a brain epidermoid cyst which shows bright signal intensity on diffusion weighted images. The patient was operated and the cystic mass removed from the preoccygeal region. Histopathological examination confirmed the diagnosis of epidermal inclusion cyst. This case report suggests that an epidermal inclusion cyst should be considered in the differential diagnosis of intractable pelvic and coccygeal pain. MRI can help to establish the correct diagnosis.

Key-word: Coccyx.
On this sequence the mass exhibited very high signal intensity related to its water restriction. This finding led us to consider that this mass could be representative of an epidermal inclusion cyst. Similar findings are also present for brain epidermoid cysts on diffusion weighted images.

Following MRI examination, the patient was operated and the cystic mass removed from the precoccygeal-presacral region. Histopathological examination of the lesion confirmed our diagnosis of epidermal inclusion cyst.

Discussion

Despite coccygeal pain being often regarded as a disease, it in fact is a symptom. Although it is mostly regarded as idiopathic or traumatic in origin, various unusual pathological conditions have been described as a cause for coccygeal pain (3, 4).

Since coccygeal pain usually develops after a local injury, trauma has been accepted widely as the aetiological factor. Antecedent trauma due to falls or difficult vaginal delivery can directly injure sacrococcygeal synchondrosis. Some rare pathological conditions which can be manifested as coccygeal pain include recent fracture (post-traumatic or intrapartum), dislocation, tumors of the sacrum and coccyx (haemangioma, carcinoid), glomus tumors of the pericoccygeal tissue, lumbosacral intradural tumors (schwannoma, ependymoma, arachnoid cysts), perineural cyst, intraosseous lipoma, infectious diseases (tuberculosis), anal duct / gland cyst, and avascular necrosis of the coccyx.

In our case report, US examination revealed a huge cystic mass in the precoccygeal – presacral region. We were able to better characterize this lesion using MRI which demonstrated the cystic nature and extension of the lesion. But the most important diagnostic clue we obtained by MRI was the high signal intensity of the lesion on diffusion weighted imaging. This finding helped us to consider an epidermal inclusion cyst in the differential diagnosis. The histopathological evaluation following surgery confirmed our diagnosis. An epidermal inclusion cyst can be described as a dermal cystic enclosure of keratinising squamous epithelium that is filled with keratin. Pathogenetically, an epidermal inclusion cyst may be secondary to congenital development or iatrogenic implantation. Clinically, these cysts manifest as painless, slow-growing, well-circumscribed swellings and may occur at any time from adolescence to adult life. They may become inflamed or secondarily infected, resulting in pain and tenderness.

In the literature, we encountered only one case report published by Jaiswal et al. (5), describing MRI findings of an epidermal inclusion cyst which presented with coccygodynia. They suggested that precoccygeal epidermal inclusion cyst should be considered as one of the differential diagnosis of coccygodynia, besides other possible aetiologies in the literature. They also mentioned that during further investigations to rule out possible aetiologies causing coccygodynia, MRI should be preferred.

Foye PM (6), commenting on the Jaiswal et al. study and highlighted the importance of MRI in detecting pathologies such as precoccygeal masses that are otherwise missed on plain radiographs. He also mentioned that physicians who routinely order lumbar or lumbosacral MRI studies for patients with coccygeal pain seem to forget to order pelvic - coccygeal MRI studies for patients with unexplained coccyx pain and thus fail to visualize the patient’s symptomatic side.

The epidermal cysts are generally characterized on MRI by the variability of signal intensity between different cases, and at times between the different parts of the same cyst (7). Other features include the absence of the oedema in surrounding tissues, fairly well-defined limits and peripheral enhancement on gadolinium ejection. The disparity in signal intensity is most likely related to the chemical state of cholesterol or the relative composition of cholesterol and keratin makes the preoperative diagnosis difficult.

Malignancy must be ruled out. Rarely, some malignancies including basal cell carcinoma, Bowen’s disease, squamous cell carcinoma and even mycosis fungoides can be developed in epidermal cysts (8).
Tokunaga et al. (9) reported a presacral epidermoid inclusion cyst in a 63 year-old Japanese man with a high CEA content. Histological examination showed that the tumor wall was made of keratinized stratified squamous epithelium without any cutaneous adnexal structure and therefore it was diagnosed as an epidermal inclusion cyst. CEA was identified in these benign epithelial cells by immunoperoxidase staining using a monoclonal antibody. This patient was the first reported case of an adult male with a presacral epidermoid cyst.

Finally, we can state that an epidermal inclusion cyst in the precoccygeal-prespacral region is an extremely rare pathology and can cause intractable pain. Correct diagnosis carries a crucial role for the treatment and MRI plays an important role for establishing a correct diagnosis. Epidermal inclusion cysts should be considered in the differential diagnosis of coccygeal pain together with other pathologies.

References