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CASE REPORT

Coccydynia with Central Sensitization plays an Important Role as Pain Generator

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ABSTRACT

Introduction: Reported is a case of "idiopathic coccydynia" with chronic pain which was refractory to conservative, medical, and interventional treatment. A diagnosis of central sensitization with coccydynia was made, and the patient responded very well to desensitization program with lignocaine and clonidine mixture. Central sensitization has proven its identity in fibromyalgia, chronic low back pain, and arthritis. But reports in coccydynia are lacking. This case report highlights the role and successful treatment of central sensitization in chronic "idiopathic coccydynia."

Keywords: Central sensitization, Coccydynia, Hyperalgesia.

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INTRODUCTION

Coccyx also known as "tailbone" is a vestigial bone that can lead to chronic pain. This pain also termed as coccydynia is still poorly understood because of the complexities in appreciating the structures and anatomy of the pelvic region, which is a site for the attachment of a multitude of muscles, membranes, and neural structures. Apart from this, the pelvic structures including the sacrum and coccyx are extensively innervated and interconnected intrinsically as well as extrinsically to the spinal cord and the muscles responsible for movements of the hip.[1]

Coccydynia has a varied etiology, such as, trauma in seated position or during childbirth, arthritis of sacroiliac joint restricting motion, spasm in muscles (levator ani, obturator internus, and pyriformis), subluxation of joints (sacroiliac, intercoccygeal, and sacrococcygeal), and load on sacrotuberous ligament, thus increasing the intercoccygeal angle (angle between first and last coccyx).[2-4] So, this multifaceted neural innervation, anatomy, and etiology make this simple entity a complex one.

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Apart from above factors, many pain physicians come across patients whose pain cannot be explained by any of the above factors, thus labeling them as "idiopathic coccydynia" linked to some psychological explanation.

Coccydynia is more common in females and is also associated with childhood abuse. As pelvic structures are said to be areas of emotional and physical stress in Indian traditional medicine, this tailbone pain becomes a social embarrassment for the patient, thus affecting their quality of life.[5] Patients usually hide their pain for long, which leads to chronicity and changes in central processing pathways causing hyperalgesia due to central sensitization. Central sensitization is postulated to be mainly due to enhanced sensitivity and excitability, loss of inhibitory control of interneurons, and reduction of inhibitory neurotransmitters.[6] In the earlier development of pain medicine, the word central sensitization was increasingly used in chronic pain syndromes causing hyperalgesia and allodynia. But there are very few data as to whether "idiopathic allodynia" can be linked to central sensitization. This case report highlights the predominant part of central sensitization in a patient with chronic tailbone pain.

CASE REPORT

A 45-year-old male patient came to our pain clinic with complaints of pain in the tailbone region for which he was taking some on-and-off analgesics for the past 4 years. He was given local steroid injection previously by his hometown pain physician. When he visited us, the pain intensity was 7 on the visual analogue score (VAS) scale. Patient complained of pain on sitting and tenderness was felt in the region of the lower sacrum and coccyx on examination. Pain was also referred to the sacroiliac region. Patient also complained of pain during defecation and sexual intercourse. Patient was subjected to some primary investigations to rule out red flags if any. Patient's coagulation profile, complete hemogram with erythrocyte sedimentation rate, and X-ray lumbosacral spine were within normal limits. After taking informed consent from the patient and ruling out absolute or relative contraindications, a caudal epidural injection was planned. An intravenous line was secured on the nondominant hand and prophylactic antibiotic was given as per hospital protocol. Standard monitoring devices were

applied. Patient was in the prone position with a pillow under the iliac crest, thus correcting the lordosis.

The legs and heels were abducted to relax the gluteal muscles, and sacral hiatus identified on lateral view. Following all aseptic precautions, 20-G epidural needle was inserted to hit the S5 vertebral body on the posterior aspect. Thereafter, the insertion angle was decreased to pierce into the sacrococcygeal membrane, thus entering into the sacral canal. The needle was further advanced upto middle of S3 in the anteroposterior view. After ruling out intravascular as well as subarachnoid injection and confirming inverted Christmas tree appearance with dye, 10 mL of lignocaine 0.5% with 40 mg depot methyl prednisolone was given therapeutically.[7] After initial pain relief for some days, the patient came back with the same pain intensity on the VAS scale. As a part of treatment, a decision to block the lowermost sympathetic ganglion (ganglion impar) was taken. Similar initial steps as above were utilized in caudal epidural block. Here, 1.5″ 22-gauge needle was introduced through the sacrococcygeal joint anterior to the anterior vertebral border. After confirming a crescent-shaped dye pattern, 40 mg depot steroid with 1% lignocaine was given.[8] Patient again returned with similar pain and was also subjected to psychological assessment. As per discussion with the psychiatrist, central role in pain causation was suspected and a diagnosis of central sensitization was made. After discussion with the patient, a 7 day desensitization program was started along with behavioral therapy. Patient was given infusion sessions each day for a week in a dose of 4 mg/kg body weight preservative-free lignocaine 2% (xylocard) in 500 mL ringer lactate with 100 µg clonidine daily over 1 hour. This desensitization program gave significant pain relief to the patient.

DISCUSSION

Pain arising from the coccyx, which was named as "the seat of the soul" by Rolf,[9] can be so devastating that it can severely affect the day-to-day activities. In 1859, pain attributed to tailbone region was given the term "coccydynia" by Simpson.[10] Although previous data and reports revealed that coccydynia is more prevalent in females, males are also not spared of this painful syndrome. It is a common condition that is self-limiting, responding to conservative management and chiropractic treatments.

A few of the unlucky cases who do not respond to treatment are difficult to manage and need psychotherapeutic treatment by the pain physicians.

Chronicity in pain medicine is a matter of debate for years due to the role of central sensitization taking place as the days pass. Central sensitization as the name suggests is sensitization and changes in the central processing pathways of pain. It was first evidenced as a part of pain hypersensitivity in 1983.[11] Typical changes include increased responses to noxious stimulation, low threshold of nociceptive specific spinal cord neurons, increased responses to stimuli on noninflamed part surrounding the inflamed area, and expansion of the receptive field.[12] Authors also called it as "spinal hyperexcitability." Overall, it can be explained as increased membrane excitability, synaptic efficacy, and a reduced inhibition.[13] This results in enhanced firing of the neurons from a segment, thus causing hyperesthesia, primary or secondary hyperalgesia, and allodynia.

Certain neurotransmitters are involved in this altered excitatory and inhibitory pathways of central sensitization, namely substance P, calcitonin gene-related peptide, vasoactive intestinal peptide, cholecystokinin, angiotensin, galanin, l-glutamate, and l-aspartate.[14] They interact with G-protein-coupled receptors on the neurons, thus rendering them hyperexcitable. Wind-up and increased firing of wide dynamic range neurons is another mechanism involved in central sensitization. It is attributed to glutamate and aspartate through N-methyl-D-aspartate (NMDA) receptor pathway. It has been revealed that excitation of this NMDA receptors induce nitric oxide (NO) synthase increasing the expression of nitric oxide. Induction of cyclooxygenase 2 in peripheral nerve lesions and enhanced release of NO and prostaglandins facilitate excitatory amino acids in the spinal cord increasing the pain intensity.[14,15]

Apart from various manual treatments available for coccydynia, simple measures of correcting the posture might help. About 90% of patients suffering from tailbone pain get relieved by simple medications, such as, nonsteroidal anti-inflammatory drugs (NSAIDs). Moreover, wedge-shaped cushions help relieve pressure while sitting.[16,17] Our patient had tried conservative treatment as well as NSAIDs/opioids but got minimal relief. Manual manipulations and massage were also tried to relieve spasms if any. So, we took the patient for interventional procedure.

Although, evidence is less for the use of interventional pain management in the treatment of coccydynia, it may be tried in chronic cases.[18] So, we employed caudal epidural block and later on sympathetic block, i.e., ganglion impar block, to block the somatic as well as the sympathetic supply in the region of the lower sacrum and the coccyx that might be responsible for pain. As the patient did not get satisfactory pain relief, a diagnosis of central sensitization with coccydynia was made. Although there is a definite role of central sensitization causing hyperalgesia in arthritis, fibromyalgia, back pain, and chronic fatigue syndrome,[19] there is no sure shot evidence of its occurrence in coccydynia. As previous conservative, medical, and interventional treatments failed in our patient, we did central desensitization program with lignocaine and

clonidine. A multitude of drugs had been tried in central desensitization, such as, pregabalin (decreases calcium influx, glutamate, substance P, and noradrenaline), gabapentin (analogue of gamma-aminobutyric acid), ketamine (NMDA antagonist), lignocaine (voltage-gated sodium channel blocker), antidepressants, such as duloxetine (activate noradrenergic descending pathways along with serotonergic pathways), tramadol (mu agonist with serotonin and noradrenaline reuptake inhibitor), tapentadol (mu opioid receptor agonist with noradrenaline reuptake inhibitor), clonidine (alpha 2 agonist and sympatholytic), and topically applied lidocaine as well as capsaicin.[20] Our patient got significant relief with central desensitization program with intravenous clonidine and lignocaine.

Psychological factors and stress response also play a role in the development of central sensitization.[21] Anxiety regarding pain, hampered daily activities as well as quality of life and associated stress lead to depression as time passes, thus starting a vicious "anxiety-pain-depression-pain" cycle. Timely assessment of our patient by the psychiatrist revealed some psychophysiological role in the causal relationship of this central sensitization and pain. All the three components (understanding role of cognition, behavior, and stresses; coping skills training using relaxation techniques, activity pacing, pleasant activity scheduling, and distraction techniques; applying and maintaining learned coping skills)[22] were taken care in cognitive behavior therapy of our patient.

CONCLUSION

Coccydynia is a common condition and is self-limited. It can be managed conservatively, but at times it can lead to central sensitization and may be difficult to treat as occurred in our case. Central sensitization was initially recognized in stroke and spinal cord injury patients, but recently, it has become a common entity in any recognizable chronic painful condition. So, a meticulous knowledge of the pathophysiologic processes in chronic pain conditions is a must so that central desensitization program can be started and targeted on pain appropriately. Apart from desensitization, a thorough psychological and behavioral rehabilitation program should be started at the earliest to help reduce suffering associated with pain and improve the quality of life.

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