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Bipolar radiofrequency ablation of the anococcygeal nerve for the treatment of chronic coccydynia: A case report

Dear Editor,

The use of radiofrequency ablation (RFA) for coccydynia has been previously described in the literature targeting various structures around the coccyx including first intercoccygeal disk, ganglion impar, and sacrococcygeal nerve. Currently, there is no consensus on which approach is most beneficial for patients with chronic coccydynia. This case describes an alternative approach targeting the anococcygeal nerve, which has not been described. A 45-year-old male presented with chronic coccyx pain after a fall. Conservative management yielded minimal improvement. To provide longer duration of symptomatic relief, bipolar RFA of the anococcygeal nerves was performed without complications and provided greater than 6 months of pain relief. Bipolar RFA of the anococcygeal nerve is an alternative method that may be a safe and effective treatment approach for coccydynia in patients who do not respond to conservative modalities. More robust research is necessary to develop a clinical framework when treating coccydynia.

Coccydynia is a condition referred to as pain in the region of the coccyx. The term was first coined by Simpson et al., in 1859. However, since then there have been limited advances in diagnostics and treatments due to complexity of the condition. Although coccydynia can affect either gender, it is five times more prevalent in women compared to men [1,3]. The female predominance of coccydynia is hypothesized to be due to a broader pelvic structure with a more posteriorly located coccyx and a risk of injury during childbirth, often accompanied by a challenging delivery [2]. Other risk factors include obesity and rapid weight loss [3]. Classically, patients present with midline pain localized to below the sacrum and above the anus; exacerbated by prolonged sitting and standing, transitioning from sitting to standing, defecation, and sexual intercourse [3]. Focal tenderness with palpation over the coccyx may be appreciated. The “Foye's finger” test has been reported in the literature where patients are asked to point to the site of most pain with one finger, similar to “Fortin's finger” test for sacroiliac joint pain [4,5]. The coccyx is a complex bony structure located at the inferior most portion of the spine that consists of three to five vertebrae that may be fused. The posterior surface of the coccyx is convex and has two prominent tubercles, known as the coccygeal cornua, which articulate with the sacral cornua of the lowest sacral segment. These cornua form the sacral foramen for the exiting sacral nerve roots. The coccyx also serves as the site of attachment for various pelvic supporting structures. Anteriorly, the coccyx is the attachment site for the levator ani, including the ischiococcygeus, iliococcygeus, and pubococcygeus muscles, and the anococcygeal ligament. On the posterior surface, the coccyx serves as one of the attachment sites for the gluteus maximus. Hence, damage to coccyx can have severe clinical and functional implications. The coccyx is primarily innervated by the sacrococcygeal plexus, which comprises of nerves originating from the ventral rami of S4, S5, and coccygeal nerve (Co1) and the dorsal rami of the sacral sympathetic trunk [6]. The coccygeal plexus gives rise to the anococcygeal nerve, which has varying anatomical description, innervation patterns, and nerve root composition. The anococcygeal nerve is composed primarily of ventral S5 and Co1 nerve roots, and in some patients the S4 nerve root [7]. After formation of the anococcygeal nerve, it penetrates through the ischiococcygeus and the sacrospinous ligament providing innervation to the cutaneous tissue at the anococcygeal region [6]. The sacrococcygeal plexus also provides variable innervation to the sacrococcygeal joint, sacrospinous and coccygeal ligament, external anal sphincter, periosteum, and the anterior musculature including the ischiococcygeus, pubococcygeus, coccygeus, and levator ani (Figs. 1 and 2) [6,8].

Etiologies of coccydynia can be classified into traumatic, non-traumatic, or idiopathic. The most common cause of coccydynia is direct, external trauma to the coccyx, such as a backwards fall which may lead to dislocation or fracture. Internal trauma primarily includes childbirth, especially difficult cases that require instrument assistance (i.e., assisted vaginal delivery with forceps). Mechanical micro-trauma can also occur from repetitive or prolonged sitting on hard surfaces. Non-traumatic coccydynia can present due to arthritic changes of the sacrum and coccyx, malignant or infectious etiologies, and variants of coccygeal morphology [3]. Idiopathic coccydynia is typically a diagnosis of exclusion that is characterized as coccygeal pain, without any gross pathologic changes involving the coccyx [9].

Coccydynia is predominately diagnosed based on clinical manifestations. A thorough history including external trauma (e.g., mechanical falls), childbirth history such as vaginal delivery with or without instrumentation assistance, weight changes, and oncological history should be obtained. A research study by Maigne et al. had found that body mass index (BMI) greater than 27.4 in women and 29.4 in men were found to be a risk factor for developing coccydynia and greater than two vaginal deliveries were associated with a higher prevalence of coccyx dislocation [10]. Physical examination should start with generalized visual inspection of overall body habitus taking in account BMI, and alignment of spine, iliac crest, sacroiliac joint, and hips. Additionally, the skin and surrounding soft tissue should be inspected for any possible masses, cysts, external hemorrhoids, anal fissures, or signs of trauma. Palpation of the posterior coccyx and surrounding soft tissue may elicit point tenderness and swelling. Pelvic and rectal examination allows for palpation of the pelvic floor musculature to appreciate for spasms or hypertonia. Additionally, coccyx manipulation may reveal hypomobility

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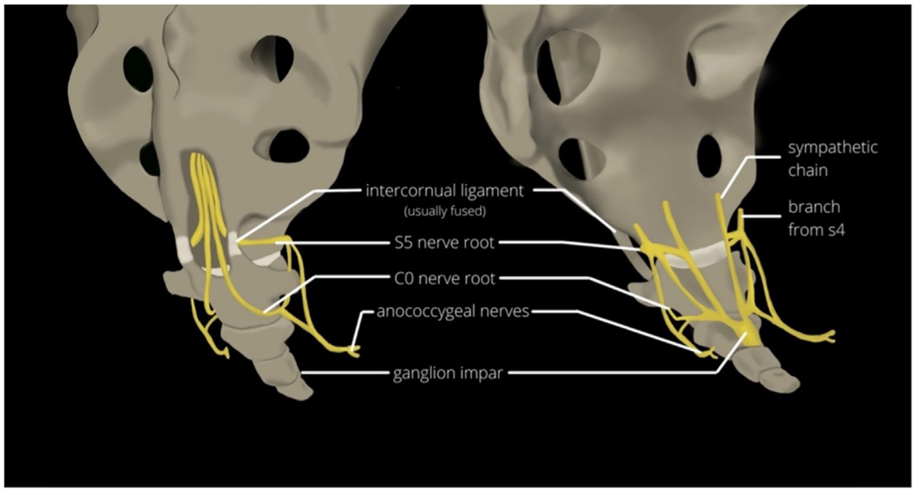

Fig. 1. Schematic demonstrating the anoccocygeal nerves and their origins and relationship to the ganglion impar.

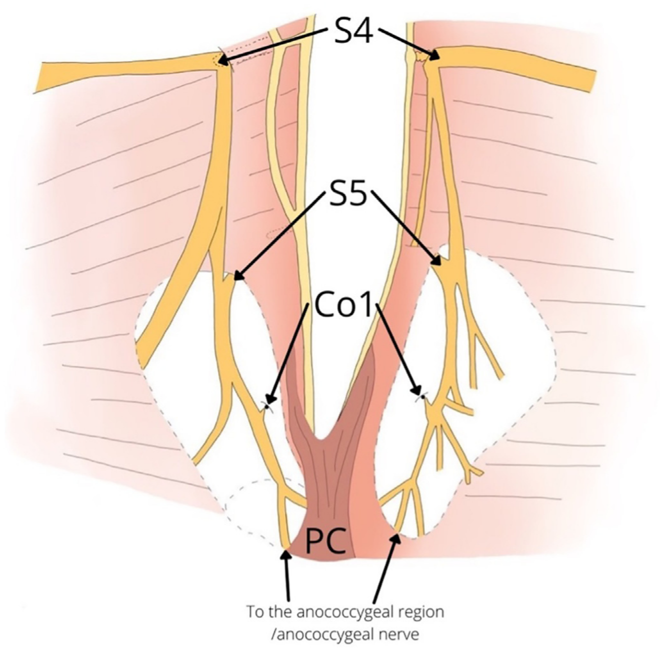

Fig. 2. Anoccoccygeal nerves along the distal lateral borders of the coccyx bones.

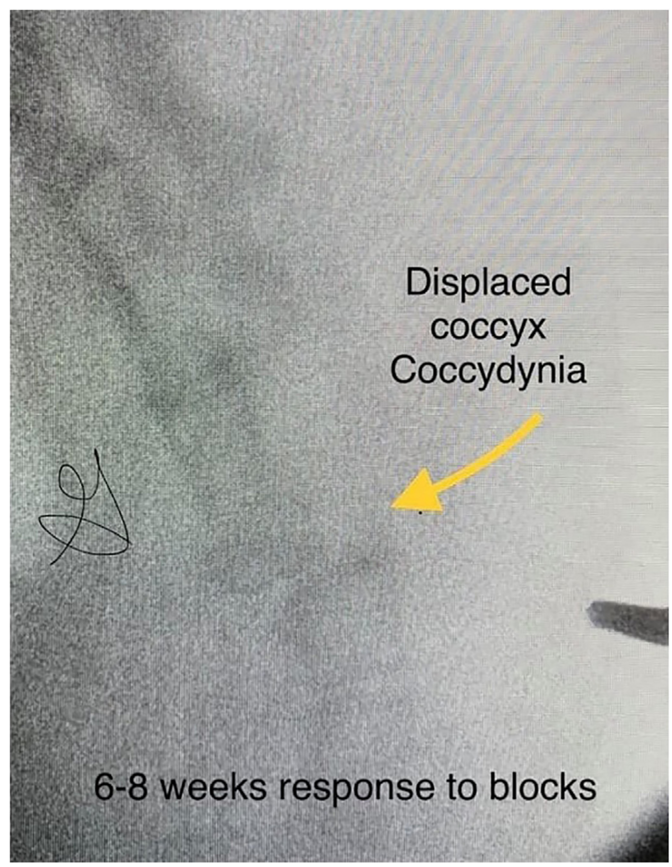

Fig. 3. Lateral fluoroscopic image demonstrating the painful, and displaced coccyx segments.

or hypermobility of the sacrococcygeal joint, as the normal range of motion of the joint is approximately 13° [3].

Although coccydynia is predominately a clinical diagnosis, radiographic studies are valuable in the evaluation process. Static radiographs provide information regarding coccyx angulation, fractures, subluxation, osteoarthritis, and osteolytic lesions. Dynamic radiographs are typically preferred to static as they provide measurements of sagittal rotation of the pelvis and coccygeal angle of incidence for evaluation of coccygeal mobility [9]. Other imaging modalities such as magnetic resonance imaging (MRI) may provide more information regarding soft tissue pathology, inflammatory processes, and cysts/masses that may cause referred pain.

Treatment varies from conservative options such as physical therapy, including pelvic floor therapy, use of wedge or donut cushions, and nonsteroidal anti-inflammatory drugs to more interventional therapies such as local anesthetic injection at the coccyx and ganglion impar block and/or neurolysis [11]. This article discusses the treatment approach with the use of radiofrequency ablation (RFA) of the anococcygeal nerve.

A 45-year-old male presented with chronic tailbone pain after sustaining a fall. X-ray imaging of the coccyx revealed displacement of the distal segment. The patient had tried conservative management including medications and physical therapy but had minimal relief. Ganglion impar blocks were ineffective, and coccygeal nerve blocks provided six to eight weeks of consistent relief (Fig. 3). In order to provide longer duration in symptomatic relief, RFA of the anococcygeal nerves was proposed.

For this procedure, 20G Stryker Venom RFA cannula and electrode

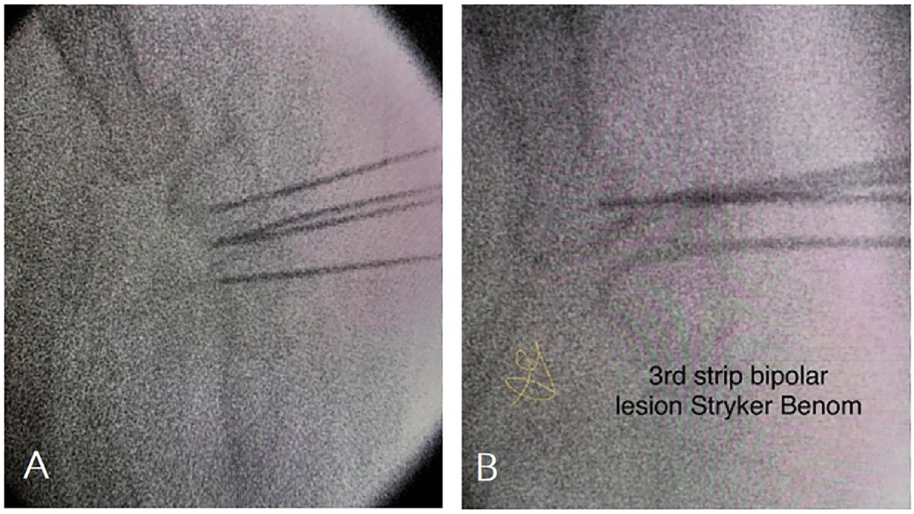

Fig. 4. Lateral fluoroscopic image of the distal coccyx. Note that it is posteriorly displaced. Bipolar radiofrequency lesioning was performed for 90 seconds at 80° Celsius, distal, and adjacent to the displaced segment.

system was used to create bipolar lesioning along the lateral aspects of the affected coccyx. Under fluoroscopic guidance, the first cannula was introduced at the lateral edge of the coccyx at the sacrococcygeal junction and advanced anteriorly, "walking off" the lateral aspect of the coccyx. A second cannula was placed approximately 10 mm caudad to the initial probe and advanced to the lateral aspect of the coccyx. A bipolar lesion was created at 80^{oC} for 90 seconds. Four sets of bipolar lesioning were performed using the "leap frog" technique, first introduced by Ferrante et al. to produce a strip lesion along the lateral edge of the coccyx to ablate the anococcygeal nerves [12]. The same technique was repeated for the contralateral side. Patient tolerated the procedure without any complications (Fig. 4).

The use of RFA in the management of chronic coccydynia has been previously described in the literature targeting various structures around the coccyx including the first intercoccygeal disk, ganglion impar, and sacrococcygeal nerve [8,13,14].

Scemama et al. reported a patient's symptoms of coccyx related pain were reproduced through a disk stimulation, confirming that the pain was through the disc and its innervation [13]. This was further supported with a local injection of anesthetics into the first coccygeal disk, which provided longer lasting relief compared to any previously tried treatments, including coccygeal steroid joint injections and bilateral coccygeal somatic nerve block. Given the improvement, the patient ultimately underwent RFA of the intercoccygeal disk and had 70% pain relief for nearly six months.

In 2014, Gopal et al. conducted a case series with twenty patients who underwent pulsed RFA treatment to the ganglion impar for coccydynia [14]. The ganglion impar is the terminal portion of the convergence of two sympathetic chains and is often targeted in the management of sympathetically mediated pain. Typically, the ganglion is positioned midline; however, there is a great deal of variability. The ganglion can be targeted through various approaches, including directly via sacrococcygeal joint or sacrococcygeal ligament, intracoccygeal approach, paracoccygeal approach, or via the anococcygeal ligament. Gopal et al. targeted the ganglion impar via the sacrococcygeal ligament and found pulsed RFA was successful in fifteen patients (75%) with reported greater than 50% improvement in pain on the visual analogue scale at 6 and 12 month follow-up [14].

Chen et al. conducted a retrospective review of twelve patients who underwent RFA of the sacrococcygeal nerves. The technique described in this report included guiding the RF cannula to the posterior lateral third regions of the coccyx and ablating from the sacrococcygeal junction to approximately the lower third of the coccyx. The number of lesions varied from two to nine and based on this technique, there was a 55.5% reduction in mean pain scores post intervention [8].

This is the first described case discussing the use of RFA targeting the anococcygeal nerve in the treatment of chronic coccydynia. The anococcygeal nerve, which is a branch of the coccygeal plexus, is primarily a sensory nerve that provides cutaneous sensation to the skin of the anococcygeal region. Fibers from the coccygeal plexus also contribute to the terminal sympathetic chains, which converge to form the ganglion impar. Although previous literature has targeted multiple structures as previously described, there is limited literature discussing targeting the anococcygeal nerve. The approach discussed in this case report is similar to the one described by Chen et al. however, the lateral border of the coccyx was targeted to capture the anococcygeal nerve as it branches off coccygeal plexus and travels along the lateral border of the coccyx.

The anatomical course and location of the anococcygeal nerve is not well defined in the literature. To overcome this, our approach utilized bipolar RF lesioning with approximately 10 mm between the RF cannula to produce a larger lesion compared to monopolar RF to effectively ablate the anococcygeal nerve. Monopolar RF produces current between the electrode tip and grounding pad. In contrast, bipolar RF transfers current between two nearby electrodes in order to produce a lesion. The size of the lesion in bipolar RF depends greatly on the inter-electrode distance, electrode tip length, and time of ablation. The early work by Pino et al. determined the upper limit of spacing between bipolar electrodes in order to produce a "strip lesion" to be 6 mm [15]. However, more recent in vivo and ex vivo work by Cosman et al. determined that 10 mm between electrodes is considered a conservative choice [16]. Although not trialed, cooled RF could theoretically be an option when targeting the anococcygeal nerve. However, clinicians need to be aware of the inherent risk of injury to surrounding soft tissue structures with larger lesion size.

Although there were no complications from this approach, the potential complications to be aware of include bleeding, infection, thermal injury, neuritis, and/or perforation of rectum [17]. In 2019, Conger et al. reported ablation related injury of the pes anserine tendon after cooled RF of the inferior medial genicular nerve [18]. Based on the close proximity to the anococcygeal nerve, there is a potential risk of ablation related injury to the ischiococcygeal and gluteus maximus muscle, as these muscles attach along the anterolateral and posterolateral border of the coccyx, respectively. Other soft tissue structures that may be potentially injured include the sacrococcygeal and sacrospinous as these ligaments have partial attachment along the lateral border of the coccyx. Careful consideration is necessary as thermoablation of tendons and ligaments can cause change in collagen structure, resulting in disruption of fibers and loss of elastic properties [19].

Another significant consideration is whether ablating the anococcygeal nerve may result in neurologic weakness or bowel/bladder incontinence. As discussed previously, the anococcygeal nerve is primarily classified as a sensory nerve that innervates the skin of the anal triangle between the distal end of the coccyx and anus. However, some studies have also described the anococcygeal nerve to supply the ischiococcygeus and part of the levator ani muscles, sacrococcygeal joint, and posterior extremity of the external anal sphincter [20–22]. Hence, the anococcygeal nerve may be involved in bowel mechanics directly via the external anal sphincter or indirectly via the pelvic floor musculature. Further studies are necessary to better understand the role of the anococcygeal nerve in bowel mechanics. Regardless, it is imperative to counsel patients of the potential risk of bowel changes with this procedure.

Coccydynia is a condition that greatly impacts the quality-of-life. Targeting the anococcygeal nerve may be an effective treatment modality in patients with chronic coccydynia that is refractory to other conservative options are. This case report adds to the growing literature of various structures that can be targeted in the treatment coccydynia, prior to more invasive surgery. Further high-quality research evaluating the long-term efficacy of these techniques and development of a treatment algorithm is necessary.

Author contributions

Concept and design: George Chang Chien, Ankur Patel, Acquisition, analysis, or interpretation of data: George Chang Chien, Daniel Wang, Ankur Patel, Illustrations: Daniel Wang, Critical revisions of the manuscript for important intellectual content: George Chang Chien, Ankur Patel, Daniel Wang, Administrative, technical, or material support: Daniel Wang, Supervision: George Chang Chien, Ankur Patel.

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