■ REVIEW ARTICLE

Coccydynia

A REVIEW OF PATHOANATOMY, AETIOLOGY, TREATMENT AND OUTCOME

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Coccydynia is a painful disorder characterised by coccygeal pain which is typically exaggerated by pressure. It remains an unsolved mystery because of the perceived unpredictability of the origin of the pain, some psychological traits that may be associated with the disorder, the presence of diverse treatment options, and varied outcomes. A more detailed classification based on the aetiology and pathoanatomy of coccydynia helps to identify patients who may benefit from conservative and surgical management.

This review focuses on the pathoanatomy, aetiology, clinical features, radiology, treatment and outcome of coccydynia.

Pain in the region of the coccyx is referred to as coccydynia or coccygodynia. A convoluted literature exists on its aetiology and management. The term coccydynia was first used by Simpson¹ in 1859, but descriptions of pain in the terminal portion of the spine date back to at least the 16th and 17th centuries.¹⁻⁴ Excision of the coccyx was carried out as early as 1726 by Jean Louis Petit.⁵ Despite being recognised for many centuries, coccydynia remains an unsolved mystery because of the perceived uncertainty in identifying the origin of the pain, some personality traits that may be associated with the disorder, the presence of a range of treatment options and varied outcomes.

Pathoanatomy

The coccyx derives its name from the ancient Greek for the similarity of the terminal portion of the spine to the beak of a cuckoo.^{3,6,7} The coccyx is a triangular bone consisting of three to five segments: the first and largest of these vertebral segments articulates with the sacrum. The last three segments diminish in size and usually form a single piece of bone. The anterior surface presents three transverse grooves indicating the lines of fusion of these segments. Attached to the anterior surface are the anterior sacrococcygeal ligaments and the fibres of the levator ani muscle. On either side of the posterior surface are the coccygeal cornua, which articulate with the cornua of the sacrum to form the posterior sacral foramen, occupied by the posterior division of the fifth sacral nerve. The flattened lateral edge of the first segment of the coccyx ascends to join the

similar border of the last sacral vertebra, thereby completing the fifth anterior sacral foramen, which accommodates the anterior division of the fifth sacral nerve. The lateral borders of the coccyx are very thin and provide attachments to the sacrosclatic ligaments, and attachments for the coccygeus muscles anterior to the ligaments and fibres of the gluteus maximus posterior to the ligaments. The tendon of the iliococcygeus muscle attaches to the tip of the coccyx, protecting the rectum and supporting it via the coccyx.

The disc spaces in this region are extremely variable. Intact discs, discs with extensive clefts, discs with cystic or fibrocystic changes and discs replaced by synovial joints have all been reported.⁸ Maigne, Guedj and Straus⁹ described the gross anatomy of the sacrococcygeal and intercoccygeal joints. In one patient the coccygeal joint had an intact disc similar to an intervertebral disc in the lumbar region, in four patients there was a synovial joint, and in a further four an intermediate structure with an extensive cleft surrounded by annular fibres or synovial cells. Balain et al⁸ reported that of 38 patients who underwent coccygectomy for coccydynia, ten had at least two discs or moving joints, and 11 had only one disc or moving joint at histology.

Aetiology of coccydynia

In Simpson's¹ classic lecture, injury to the coccyx or coccygeal joints with surrounding tissue inflammation and contraction of the muscles attached to the coccyx causes the characteristic pain of coccydynia. Even though

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Table I. Classification of coccydynia based on aetiology

A:	Based on aetiology
	1. Idiopathic
	2. Traumatic
B:	Based on pathology
	1. Degeneration of the sacrococcygeal and intercoccygeal disc and joints
	2. Morphology of the coccyx: type II, III, IV, presence of a bony spicule and coccygeal retroversion
	3. Mobility of the coccyx: hypermobile or posterior subluxation
	4. Referred pain: lumbar pathology or arachnoiditis of the sacral nerve roots, spasm of the pelvic floor muscles and inflammation of the pericoccygeal soft tissues
	5. Others: neoplasm, crystal deposits, infections
C:	Somatisation or neurotic

Table II. Modification of Postacchini and Massobrio classification¹³ based on morphology of the coccyx

Type	Coccygeal morphology
I	Curved gently forward
II	Has a marked curve with the apex pointing straight forward
III	Angled forward sharply between first and second or second and third segments
IV	Anteriorly subluxated at the level of the sacrococcygeal joint or first or second intercoccygeal joint
V	Coccygeal retroversion with spicule
VI	Scoliotic deformity

Simpson's statement still holds true, the factors that provoke the pain appear diverse. Traycoff, Crayton and Dodson¹⁰ classified coccygeal pain as either pain that arises from the coccyx itself, pain that is referred to the coccyx, or pain that is neurogenic in origin, arising from the nerve roots, plexus or peripheral nerves that travel to or through the coccyx. However, his classification is limited. A more detailed classification should not only be based on the site of pain, but should also encompass the pathoanatomy of the origin of pain (Table I).

The most common cause of coccydynia is single direct axial trauma such as a fall directly onto the coccyx or, as during the post-partum period, due to a subtle form of cumulative trauma that occurs due to sitting awkwardly.¹¹ Schapiro,¹¹ in 1950, described the disorder as 'television disease', because poor postural adaptation was thought to be an important predisposing factor of coccydynia. Maigne et al⁹ reported that 36 of 51 patients had a history of direct trauma, and Pennekamp et al⁷ reported a 50% incidence of direct trauma. Depending on the severity of the trauma, patients experience strain to the pelvic floor muscles, mild distortion without bony or ligamentous damage over a fissure in the caudal coccygeal segments, or a severely dislocated fracture of the sacrococcygeal complex.⁷ Maigne, Doursounian and Chatellier¹² reported a body mass index of more than 27.4 in women and 29.4 in men as a risk factor for the development of both idiopathic and post-traumatic coccydynia. When obese individuals attempt to sit down, the coccyx tends to jut out posteriorly as a result

of inadequate sagittal pelvic rotation. This results in increased exposure to the intrapelvic pressure that develops during the act of sitting or during a fall, resulting in subluxation of the coccyx.¹²

Postacchini and Massobrio¹³ suggested that the morphology of the coccyx may have a role in the aetiology of coccydynia. We have modified their classification to incorporate retroversion and scoliosis of the coccyx (Table II). Based on the classification of morphology, type II, III and IV patients were at higher risk of developing coccydynia. Dennell and Nathan¹⁴ reported a case of coccygeal retroversion with a small palpable bony excrescence on the dorsal aspect of the coccyx, termed a spicule, along with the presence of a small pilonidal sinus pit. The spicule was identified in 14% of 208 coccydynia patients by Maigne et al,¹² who reported an anterior subluxation of the coccyx. According to Maigne et al,¹² spicules cause irritation of the coccygeal region when the patient is sitting, especially in immobile coccyges, which results in the development of chronic adventitial bursitis. Marmor¹⁵ expressed the opinion that coccydynia was due to coccygitis, a condition similar to epicondylitis of the elbow. Kim and Suk⁴ reported the presence of scoliotic deformity of the coccyx as a possible cause of coccydynia.

Maigne et al⁹ classified coccygeal mobility into four groups: luxation-backward displacement of the mobile portion of the coccyx when the patient is sitting; hypermobility, where coccygeal flexion exceeds 25° when the patient is sitting; immobile coccyx with < 5° of flexion or

extension when the patient is sitting; and normal mobility, where coccygeal mobility is between 5° and 25°. Luxation and hypermobility are abnormal by themselves, and the immobile coccyx associated with spicules due to the formation of bursa is an anatomical cause of coccydynia.

Owing to the presence of discs or disc-like structures in gross anatomical specimens, a degenerative cause of pain has been considered. Bayne, Bateman and Cameron¹⁶ were the first to report that 41% of patients with idiopathic coccydynia and 44% with traumatic coccydynia had objective evidence of degeneration. According to Alo, Eisenstein and Darby,¹⁷ five of the eight patients who had a coccygectomy for coccydynia had histological evidence of disc degeneration, two had degenerative articular cartilage changes, and one had a near-normal disc. In a review article, Fogel, Cunningham and Esses¹⁸ noted no pathological findings in patients with coccydynia. Wray, Easom and Hoskinson¹⁹ also reported that histological examination of the coccyx had not helped in revealing the cause of coccydynia.

Maigne et al⁹ suggested that common coccygeal pain may be discogenic in origin, based on findings of provocative discography. In his series, 15 of 21 patients yielded a positive result on provocative discography, and 25% had coccygeal luxation in the sitting position which was reducible in other positions. The presence of dynamic instability indicates that the cause of pain is probably the result of degeneration, as in other regions of the spine. However, unlike lumbar discs, coccygeal discs do not develop osteophytes/syndesmophytes because they do not sustain compressive loads.

Pre-existing back pain or coccydynia as a manifestation of lumbar disc pathology has been addressed in detail in the literature. Postacchini and Massobrio¹³ reported that 31% of their patients had associated low back pain. According to Bayne et al,¹⁶ 15% of their patients presented with pre-existing back pain, and Perkins, Schofferman and Reynolds²⁰ reported that 77% of patients had co-existing lumbar spine disorders. Balain et al⁸ reported a 71% incidence of concomitant lumbar spine disorders.

Various rare pathological lesions have been characterised as the cause of coccydynia. The first reported coccygectomy was undertaken in 1726 for tuberculosis.⁵ Richette, Maigne and Bardin²¹ described the deposition of calcium crystals in the sacrococcygeal or intercoccygeal joints as a cause of coccydynia. Tumours such as chordoma, trichoblastic carcinoma and notochordal cell tumours have also been reported.

The dilemma that the origin of pain could not be objectively evaluated led to the suggestion that patients with coccydynia were neurotic. Balain et al⁸ reported that coccydynia had been under-represented, especially as the motivation of some patients was suspect. Bremer²² was the first to describe the association of neurosis with coccydynia. Dittrich²³ described the presence of myofascial trigger points over the sacrococcygeal fat as the cause of pain. In 1959, Smith²⁴ described a separate group of patients with

psychogenic disorders such as neurosis and hysteria as a cause of coccydynia. Even though a psychological basis cannot be completely ruled out, most cases of coccydynia require careful evaluation of the cause, and neurosis should be an aetiology of exclusion.

Clinical features

Coccydynia constitutes less than 1% of all non-traumatic complaints of the spine.^{7,25} Women are five times more commonly affected than men.^{7,25} In 1950, Schapiro¹¹ confirmed the findings of Thiele²⁵ on the clinical features of coccydynia and called it Thiele syndrome. Thiele's description²⁵ of the clinical features still holds true today. This includes the main symptoms of tenderness and pain, or an ache localised in the region of the lower sacrum, the coccyx, or in the adjacent muscles and soft tissues. The patient usually points to the coccyx as the site of pain. The severity of the pain is dependent on various predisposing factors, such as the duration of time spent sitting. Women report an exponential increase in pain during the premenstrual period. Dyspareunia and piriformis syndrome have been infrequently associated with coccydynia. The character of the pain appears to be more related to spasm of the levator muscle, as patients complain of pain during defecation or sexual intercourse.

Radiological features

The diagnosis of coccydynia was based predominantly on clinical examination with static standard radiographs, CT, and routine blood tests, which showed no particular abnormality except in the presence of tumour or infection. Coccydynia is a dynamic disorder which can only be appreciated on dynamic films.

Maigne et al⁹ described a method of assessing coccygeal mobility based on a comparison of static and dynamic lateral films with the hips flexed while the patient sat on a hard stool and extended the spine, producing more pain. The films were superimposed to compare and measure the sagittal movement of the coccyx in flexion and extension. The angles of sagittal pelvic rotation, the angle of mobility and the angle of incidence were calculated. The angle of sagittal rotation was measured based on the pivot angle made when the radiographs were superimposed, matching the sacrum. The angle of incidence measured the angle at which the coccyx struck the seat surface. The angle of mobility was the difference between the tips of the coccygeal segments in the two films. Based on the mobility of the coccyx, it was classified as normal, subluxed, immobile or hypermobile. Kim and Suk⁴ assessed the radiological differences between idiopathic and traumatic causes of coccydynia based on the description of the intercoccygeal angle, and the angle between the first and last segments of the coccyx, which was greater in patients with idiopathic coccydynia.

Provocative discography has been found to be a more promising investigation in the assessment of coccydynia. Maigne et al⁹ were the first to describe coccygeal discogra-

phy, a procedure similar to lumbar discography. Based on the objective evidence of degeneration as a cause of pain, assessment of the disc space using a discogram to reproduce the pain appears to be a more valid evaluation tool. Maigne et al⁹ reported that coccygeal discography succeeded technically in 44 of 51 cases, and was positive in all cases with subluxation or hypermobility. Half of the patients in this investigation with normal dynamic films had a positive discogram. However, Balain et al⁸ reported that five of the six patients who had discograms had some clinical findings, although they could not identify the painful levels on discography, nor was it significant in identifying suitable candidates for surgery.

Static MRI failed to identify the cause of coccydynia, as it is a dynamic disorder. Grassi et al²⁶ reported the use of dynamic MRI to indicate that the coccyx is mobile during defecation, and were able to demonstrate coccygeal excursion by assessing the difference between its position at maximum contraction and during straining or evacuation.

Treatment options

Many different treatment methods have been proposed for the management of coccydynia. The spectrum ranges from simple measures such as the use of laxatives, non-steroidal anti-inflammatory drugs, hot baths, ring-shaped cushions, ergonomic adaptations to physical therapy, intrarectal massage and manipulation, sacrococcygeal injections, ganglion impar blocks, radiofrequency thermocoagulation, psychotherapy, and coccygeoplasty to coccygectomy (partial or complete). Conservative management is successful in approximately 90% of patients.^{25,27,28}

Ergonomic adaptation is commonly used. Methods include strapping of the buttocks, postural training and the use of a rubber ring or a firm corset, but there is little evidence to support their use. Based on the pathophysiology, pain is predominantly due to spasm of the pelvic floor muscles. Massage and manipulation therapy are also often used. Thiele²⁵ described a vicious cycle in the production of pain in the pelvic floor. Following an inciting factor, spasm occurs in the muscles of the pelvic floor, the levators and the coccygeus. One must remember that muscle spasm by itself is a painful condition. Spasm of both portions of the levator produce forward as well as lateral traction of the coccygeus muscle. Unilateral contraction of the coccygeus muscle pulls the coccyx to one side, and in the presence of coexisting sacrococcygeal joint pain or degeneration the pain tends to increase, resulting in more spasm. Based on the hypothesis of the pathophysiology of pain, Thiele²⁵ described massage of the levator muscles along the long direction of the fibres on both sides.

Maigne et al²⁹ reported that massage and levator stretch as described by Thiele was intended to treat pain with tonic spasm. However, in patients in whom pain was due to mobility, Thiele's manoeuvre was not helpful, but mobilisation of the coccyx might be. Mennell³⁰ described a technique of manual manipulation in which the coccyx is grasped

between the thumb and the index finger inserted into the anal canal and manipulated. Maigne et al²⁹ also advocated an addition to the above technique whereby, following manipulation, the coccyx is maintained in hyperextension with the index finger applied to the ventral aspect of the inferior sacrum while counter-pressure is exerted by the left hand over the posterior sacrum. These authors proposed a new type of manipulation for patients who had restricted sacrococcygeal extension, in which the coccygeus, levator and external sphincter are stretched by keeping the coccyx still with one finger internal and one external.

Injections using steroid or local anaesthetic have become popular, both as treatment and as a tool to assess patients who would benefit from coccygectomy. Although injections into the pericoccygeal tissues have had a major impact, there is no clear consensus in the literature regarding the site of injection. Plancarte et al³¹ first described the use of radiofrequency ganglion impar blocks for pericoccygeal pain due to carcinoma. Recent studies^{32,33} have used this technique to alleviate pain in patients with severe coccydynia.

Coccygectomy (the surgical removal of the coccyx) is used in patients who do not respond to conservative measures. Powers⁵ stated that coccygectomy fell into disrepute because it was used inappropriately. Two techniques of coccygectomy have been discussed in the literature. Powers⁵ technique positions the patient prone with the buttocks firmly taped laterally for ease of exposure of the coccyx. A midline vertical incision over the coccyx is developed down to the bone, and includes exposure of the sacrococcygeal joint. The dissection is carried distally to expose the tip of the coccyx. A subperiosteal plane is developed on the anterior surface, taking great care to stay clear of the rectum. The sacrum is amputated just proximal to the sacrococcygeal joint, and the entire coccyx with the sacrococcygeal joints is lifted free and excised. Haemostasis is achieved and the wound closed in layers.

Gardner³⁴ described an improved technique which does not involve the most hazardous step of Powers' approach of placing a finger in the rectum, which leads to increased risk of infection and the development of a blind plane for the rectum which may result in rectal injury. The technique is based on a modification of the Kraske procedure for treating cancer of the rectum with preservation of the anal sphincter.^{5,34} According to Gardner,³⁴ a 7.5 cm incision extends from just proximal to the sacrococcygeal joint into the buttock crease, care being taken to prevent it extending into the peri-anal region. The incision is brought down to the fascia and insertion of the gluteus maximus directly over the bone. The coccygeus vessels are ligated. The tip of the coccyx is elevated by blunt dissection. Then, the tip of the coccyx is separated from the external sphincter by sharp dissection. The coccyx can then be easily elevated from the underlying rectum and the dense fascia that separates the two. Using a moist sponge, the fascia and the rectum are mobilised away from the coccyx proximally up to the sacrococcygeal joint. The haemorrhoidal vessels are ligated under

Table III. Outcome of coccygectomy for coccydynia

Authors	Number of patients	Number of excellent to good results (%)
Postacchini and Massobrio ¹³	36	32 (89)
Hellberg and Strange-Vognsen ⁴⁰	55	46 (83)
Wray et al ¹⁹	23	21 (91)
Grosso and van Dam ²	9	8 (89)
Perkins et al ²⁰	13	12 (92)
Shoakazemi et al ⁴²	13	10 (77)
Trollegaard et al ²⁸	41	33 (81)

direct vision, and the coccyx is excised by sharp dissection at the sacrococcygeal joint. The advantages of this technique include removal of the coccyx *in toto*; less risk of damage to the adjoining structures such as the rectum, removing the necessity for placing the finger in the rectum; and less traumatic dissection, leading to less pain and early recovery.

Dean et al³⁵ reported injection of polymethylmethacrylate cement in a patient with a coccygeal fracture which resulted in immediate relief of pain. They termed the procedure a coccygeoplasty. In another report, Bergkamp and Verhaar³⁶ reported the use of vicryl sutures as a tension band in a patient with coccydynia and dislocation of the coccyx to maintain the integrity of the coccyx.

Functional outcomes

Conservative treatment remains the mainstay of treatment. There is wide variation in the results of injection techniques. Wray et al¹⁹ reported 60% improvement with pericoccygeal injection, 85% with manipulation and injection and 90% with coccygectomy. Ramsey et al³⁷ reported a 78% success rate for manipulation and injection, and a 87% success rate with surgery. Perkins et al²⁰ reported that 75% had relief after injection and 92% after surgery. A few authors^{8,38} have commented on the role of steroid injection as a pre-operative test to assess the efficacy of surgery. Balain et al⁸ reported a 59% surgical success rate in non-responders following injection, compared with 75% in responders. Table III summarises the literature on coccygectomy and the outcomes of various studies.

Powers⁵ and Key³⁹ felt that a two-month trial of conservative treatment was sufficient for patients who were non-responders, whereas others^{4,13,40,41} recommended three to eight months as an ideal waiting time to determine the outcome of conservative methods.

Various authors^{2,13,19,20,28,40,42} have reported good to excellent results in patients who underwent coccygectomy for coccydynia (Table III). Careful patient selection appears to be the key to success. Various clinical and radiological pointers predict a good outcome after surgery. Coccygectomy for coccydynia due to changes in the shape of the coccyx, the presence of a bursa, and a good response to injection has an increased chance of an excellent result after surgery.

Infection remains the most important complication of coccygectomy because of the proximity of the incision to

the rectum and anal canal. Bayne et al¹⁶ reported an infection rate of 16.6%, and Doursounian et al⁴³ reported a 14.75% rate. Postacchini and Massobrio¹³ reported partial skin necrosis and superficial wound infection causing delayed healing in up to 50% of patients. Balain et al⁸ reported that the use of prophylactic antibiotics substantially reduced the risk of infection. Modification of the Kraske technique as described by Powers⁵ also substantially reduces the incidence of infection. Complications such as rectal injury, anal sphincter injury and incontinence have been rarely reported.

Conservative treatment in the form of massage, injection and ganglion impar blocks appear to play a vital role in the management of coccydynia. Coccygectomy is effective in patients with coccydynia, especially for those with specific indications. Even though there appears to be a psychological issue associated with diagnosis, there are studies that report good or excellent results for patients with the appropriate indications undergoing coccygectomy.

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