Coccygectomy for treatment of coccydynia: A systematic review of literature

Submitted in partial fulfillment of Master Degree

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Finally, I hope that this review to add value to the pool of science and the world of research.

Aim of the work

The aim of this work is to do a systematic article review of English literature on the topic of coccygectomy for treatment of coccydynia.

Contents

Coccygectomy for treatment of coccydynia: A systematic review of literature

Abstract

Coccydynia is a pathological condition associated with pain discomfort all around the bottom end of the spine. The aetiology and the intensity of the symptoms may defer significantly. The effectiveness of the surgical treatment remains obscure. Our purpose, through this systematic review is to evaluate the results of surgical treatment of coccygectomy. Literature retrieval was performed by the use of the PubMed searching engine utilizing the terms 'coccydynia–coccygectomy' in the English language from January 1980 to January 2017.

Case reports and tumour related case series were excluded as well as articles published in other languages. In total 36 manuscripts were analyzed. Only 4 of them were prospective studies whereas 32 were retrospective case series; seven were classified as Level III studies and the remaining as Level IV studies. In total, 1040 patients with coccydynia underwent coccygectomy following failed conservative management. The sex ratio, male/female was (223:1028) 1:4.6.

The most popular etiology for coccydynia was direct trauma in 538 patients. 806 of the patients reported an excellent/ good outcome following the procedure. There were 9 deep and 76 superficial infections. Other complications included 2 hematomas, 6 delayed wound healings, 3 persistent drainage and 13 wound dehiscence.

The overall complication rate was 11.5%. Patients with history of spinal or rectal disorders, as well as idiopathic or with compensation issues, had less predictable outcome than those with history of trauma or child- birth. Coccygectomy can provide pain relief to as high as 84.6% of the cases. The most common reported complication was wound infection.

Introduction

Coccydynia was initially described by Simpson [1] as pain–discomfort localized around the bottom end of the spine and usually triggered by prolonged sitting. The aetiology and the intensity of the symptoms may defer significantly. It could be the result of severe trauma, repetitive injury, post childbirth, local tumours, disc degeneration and of idiopathic nature [2–6, 34]. The exact pathophysiologic mechanisms associated with coccydynia are still obscure, while obesity is considered as a predisposing factor due to the resulting pelvic rotation [2].

Coccydynia has been shown to be more common in the female population. This has been attributed to specific sex-related anatomic features (slender females have little subcutaneous fat and the coccyx is relatively unpadded; greater sciatic notch being wider, contributes to backward inclination of the sacrum and coccyx, thus increasing the possibility of injury to the area of the coccyx; females have greater inter-ischial tuberosity distance which increases the pressure to the coccyx) [3, 4]. Furthermore, it has been reported that psychological depression, neurosis or hysteria, magnifies the clinical symptoms [5, 6].

Table 1 The table provides detailed information regarding the Postacchini and Massobrio [7]

Coccygeal configuration types I, II, III, and IV (from left to right). Type I: Coccyx is curved slightly forward, with its apex directed downward and caudally. Type II: Forward curvature is more marked, and apex extends straight forward. Type III: Coccyx most sharply angulates forward. Type IV: Coccyx is subluxated at sacrococcygeal joint or at intercoccygeal joint.

Maigne et al. [2, 8, 9]
introduced the use of dynamic
X-rays, comparing standing
and sitting radiographs Figure (1)
Type I: Curving more than 25° Figure (2)
Type II: Displaced or sublaxated
Posteriorly Figure (3b)
Type III: Immobile with a spicule
in the dorsal surface of the last
coccygeal segment. Figure (4)

Figure 1. *The correct position to X-ray the coccyx in a sitting posture. Note the foot rest.*

Figure 2. Coccygeal flexion greater than 25° in the sitting position

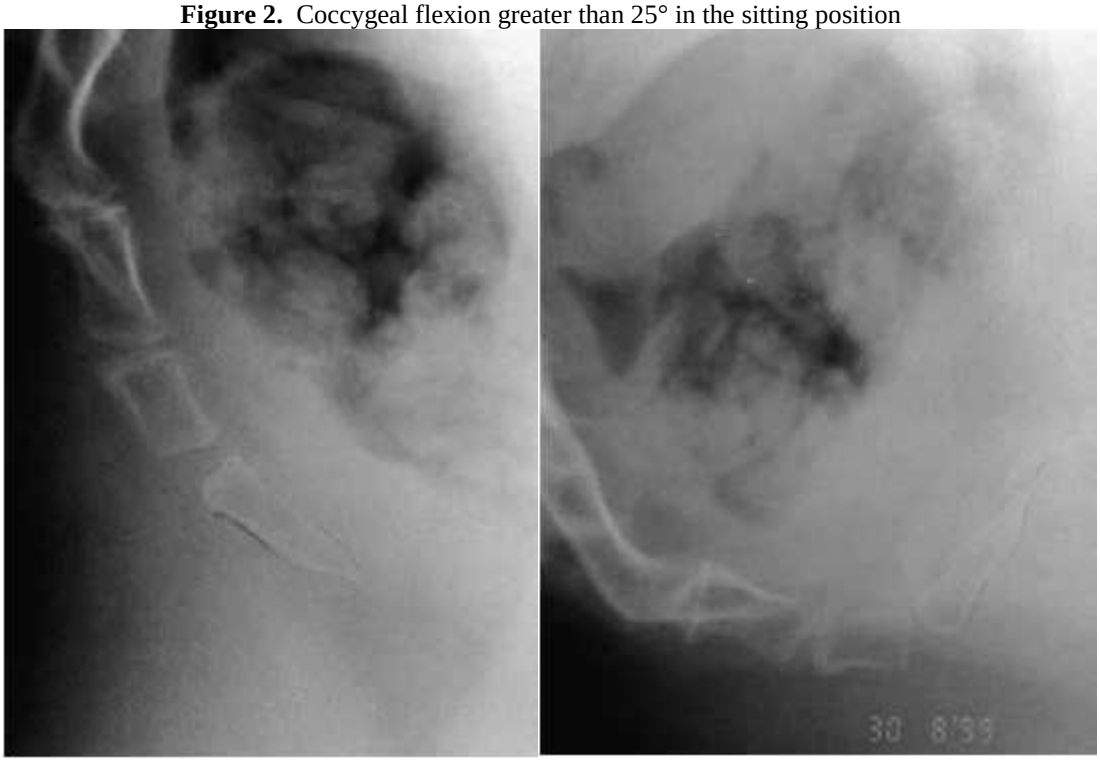
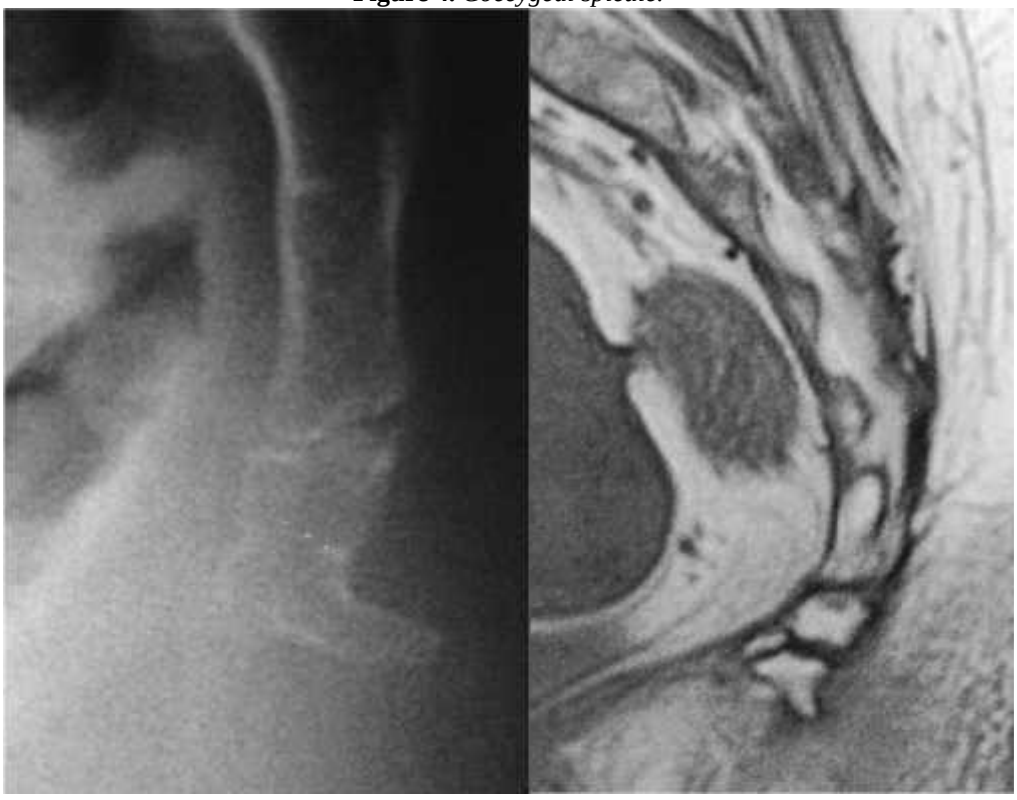


Figure 3. Radiographs showing a) the normal pattern of the coccyx in the standing subject and b) subluxation when the subject is sitting.

Figure 4. *Coccygeal spicule.*



Both clinical signs (pain and abnormal movement on palpation) and radiological features can aid in making the diagnosis. Radiological evaluation is usually performed with lateral sacral radiographs, allowing classification of the shape of the coccyx according to its inclination [7] (Table 1). The use of dynamic X-rays, comparing standing and sitting radiographs has also been found useful [2, 8, 9] (Table 1). MRI and bone scintigraphy can provide valuable information excluding the presence of degenerative spine disease, local tumour pathology or metastatic bone disease.

Treatment options of coccydynia can be non-operative or operative. Non-operative treatment includes rest, NSAID's drugs, doughnut-shape cushion usage, physiotherapy, radiotherapy, sacral rhizotomy, epidural injections and local injections with or without coccyx manipulation. In cases of persistent symptoms unresponsive to conservative treatment, coccygectomy is offered as the definitive surgical solution. Two different types of surgical resection have been described (Table 2), with the most popular one being the procedure introduced by Key [10], which differs from that of Gardner [11], as to the direction of the coccyx dissection from the rectum. According to Postacchini and Massobrio [7], the coccyx can be removed either totally or partially with comparable good results. A sub-periosteal resection instead of a total coccygectomy has also been reported as an option [12]. However, the literature regarding the results of surgical treatment is vague. The purpose of this study therefore was to evaluate the results and complications, as well as the patient's satisfaction following surgical treatment of coccydynia (coccygectomy).

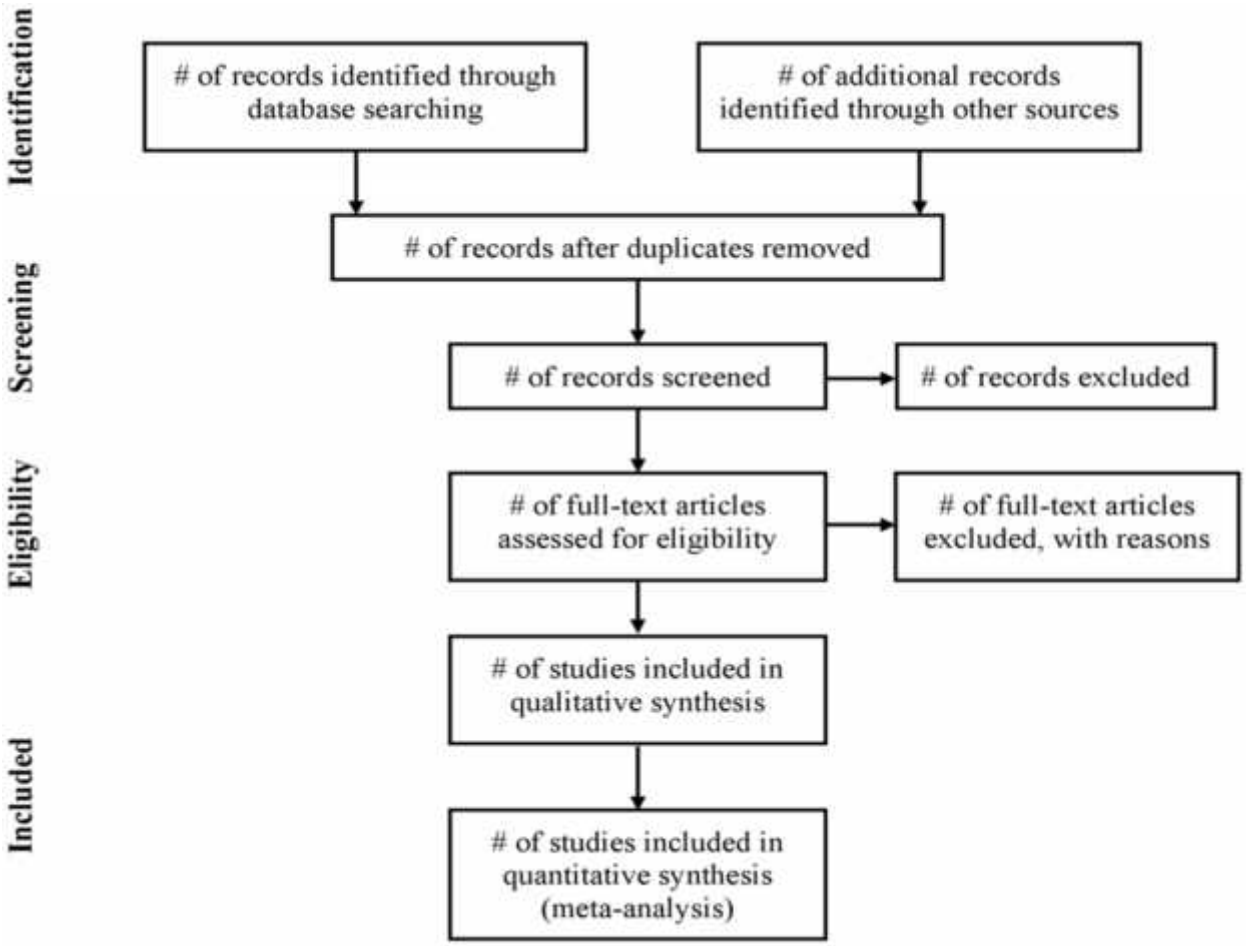
Table 2 Coccygectomy surgical suggestion

Key [10]	Coccygectomy performed from proximal to Distal
Gardner [11]	Coccygectomy performed from distal (tip) to proximal with the advocate disadvantage that the surgeon works blindly and that increases the risk of rectal injuries
Postacchini and Massobrio [7]	Coccyx can be removed either totally or partially with comparable good results
Bilgic et al. [12]	Prevalence of the sub-periosteal resection compared with coccygectomy

Materials and methods

Literature retrieval was performed by the use of PubMed Searching machine and by using the terms ‘‘coccydynia’’ and/or ‘‘coccygectomy’’ as key words. The searching limits included English literature and Human studies. Eighty-five articles were initially identified. Case reports, editorials and tumour-related case series were excluded, as well as case series referring solely to other methods of management and not surgical resection. Series comparing surgical versus injection therapy were included. Further research was made through the articles references.

All extracted articles did not meet the selection criteria, as they didn't mention the surgical option and outcomes or were a case reports .Manuscripts selected were analyzed according to the data providing for the number of patients, including age, gender characteristics, the period of pre-exciting symptoms, the aetiology and classification, the type of surgery, the use of antibiotics or drain, the follow-up period and the frequency of perioperative complications.



Eligibility criteria:

Inclusion criteria:

Studies are eligible for inclusion if they meet the following criteria:

- Series of coccygectomy for coccydynia
- Series reporting surgical outcomes.
- Journal articles only
- English literature only
- Human studies only

Exclusion criteria:

- Studies that did not report surgical option for coccydynia
- Series not reporting surgical outcomes.
- Animal studies.
- Case reports.
- Tumour-related case series

Data analysis:

Data from different studies will be compared when similar criteria are used to assess surgical outcomes and also to report complications. We examined the full texts of the potential studies before finalizing their inclusion in the review.

Results

In total 36 manuscripts met the inclusion criteria and were analyzed (Tables 3, 4, 5) [13–32]. Only four of them [4, 44, 18, 51] were prospective series, and seven were classified as Level III studies [4, 12, 18, 23, 26, 44, 51]. The majority were retrospective uncontrolled case series. In total, the reviewed series included 1040 patients with coccydynia that had coccygectomy as definitive pain management.

Table 3 Systematic review data, providing paper characteristics, materials, duration of symptoms, aetiology and classification

Authors	Study	Evidence	Patients	Females/ males	Age	Range	Duration of symptoms	Etiology	classification
Trollegaard[41]	Retro	IV	41	39/2	39.1	(16–77)	6 months	21 trauma ,8 childbirth, 12 idiopathic	n/s
Shao-wen [42]	Retro	IV	31	26/5	41.5	(20–65)	9.6 months	12 trauma,3 idiopathic , 16 congenital	n/s
Khorrami [43]	Retro	IV	13	10/3	35.5	(6–57)	4-6months	9 trauma, 4 idiopathic	n/s
Hanley [44]	Pro	III	98	87/11	47.2	(19–76)	5 years	35 trauma,47 idiopathic,7 childbirth,8 wt loss	Maigne
Jacquot [45]	Retro	IV	33	31/2	42	(23–62)	n/s	33 Coccygeal spicules,3 of them wt loss	Maigne
Ramieri [46]	Retro	IV	31	19/12	31	(21–47)	33 months	31 trauma	n/s
Haddad [47]	Retro	IV	14	12/2	39.4	(21–47)	30 months	8 trauma,3 idiopathic,1 childbirth,2 post fusion	
Awwad [48]	Retro	IV	70/8 oper	52/18	42	(16–58)	6 months	34 trauma, 14 childbirth,22 idiopathic	n/s
Ogur [49]	Retro	IV	22	15/7	33.6	(23–46)	1 year	22 trauma	Postacchini
Wood and Greger [50]	Retro	IV	40	32/8	-----	-------	2 years	18 trauma,22 idiopathic	Postacchini and Maigne
Wray and Templeton [13]	Retro	IV	37	32/5	34	(17–53)	4 years	n/s	n/s
Postacchini and Massobrio [7]	Retro	IV	51	44/7	34	(11–44)	(2 months–15 years)	32 trauma, 19 idiopathic	Postacchini
Bayne et al. [5]	Retro	IV	48	38/10	39	(14–71)	n/s	25 trauma/12 idiopathic/7 spinal operation/4	n/s
Eng et al. [14]	Retro	IV	27	25/2	34.5	(13–74)	n/s	16 trauma, 1 child, 1 inject	n/s
Hellberg [15]	Retro	IV	65	58/7	34	(12–65)	6 months	32 trauma/8 childbirth/25 idiopathic	n/s
Wray et al. [4]	Pro	III	120//23 oper	101/19	38 f/47 m	(11–74)/ (13–76)	16 months (6–10 years)	30 trauma /14 childbirth/15 injury	n/s

Grosso and van Dam [16]	Retro	IV	9	6/3	32.5	55months	(7 months–14 years)	6 trauma/3 idiopathic	Postacchini
Zayer [17]	Retro	IV	10	10/0	46	(26–74)	n/s	5 trauma/2 child/1 surgery/2 idiopathic	n/s
Maigne et al. [18]	Pro	III	37	28/9	46.5	(30–64)	31 months	n/s	Maigne
Perkins et al. [19]	Retro	IV	13	9/4	45	(24–72)	32 months (9–44 months)	7 trauma/6 idiop	n/s
Ramsey et al. [20]	Retro	IV	24 inj/15 sur	14/1	32	n/s	n/s	5 trauma/8 idiopathic/1 childbirth	n/s
Doursounian et al. [21]	Retro	IV	61	49/12	45.3	(18–72)	30 (2–28)	n/s	Maigne
Hodges et al. [22]	Retro	IV	32/11 oper	24/8	47	(23–72)	6 months	3 childbirth/15 trauma 14 idiopathic/	n/s
Wood and Mehbod [23]	Retro	III	20 oper /25 inj	41/10	41	(22–66)	22 (14–47)	28 trauma/6 childbirth/17 idiopathic	Postacchini
Karalezli et al. [24]	Retro	IV	14	14/0	28	(17–39)	n/ s	8 trauma/3 childbirth/3 idiop	Postacchini
Feldbrin et al. [25]	Retro	IV	9	7/2	52.8	(21–65)	14 (4 months–2 years)	4 trauma/2 idiopathic/2 childbirth/1 meta	Postacchini
Pennekamp et al. [26]	Retro	III	16	14/2	39.8	(11–75)	n/s	8 trauma/8 idiopathic	n/s
Balain et al. [27]	Retro	IV	38	31/7	47.3	(15–71)	n/s	15 trauma/3 childbirth/13 idiopathic	n/s
Mouhsine et al. [28]	Retro	IV	15	9/6	44.2	(33–58)	6 months	Trauma all	Maigne
Capar et al. [29]	Retro	IV	24	23/1	33	(21–60)	30 (14–144)	18 trauma/6 idiopathic	Postacchini
Sehirlioglou et al. [30]	Retro	IV	74	64/10	43.4	(16–45)	7 (3–12 months)	n/s	n/s
Cebesoy et al. [31]	Retro	IV	21	15/6	31	(28–39)	8 months (6–10 months)	19 trauma/2 childbirth	n/s
Bilgic et al. [12]	Retro	III	25	15/10	26.4	(20–39)	10 (8–13)	17 trauma/8 instability	n/s
Traub et al. [32]	Retro	IV	8	6/2	47.3	n/s	15.2 months	5 trauma/3 idiopathic	n/s
Arafa et al. [51]	Pro	III	38	28/10	41.1	(29–55)	19.6 months (9-48)	Trauma all	Postacchini and Maigne

The range of the mean ages of those series was from 26.4 [12] to 52.8 years [25]. The extremes of the age range were 6 [43] and 77 [41] years.

Gender characteristics were provided on 963 patients. The vast majority of the patients were females (796, 82.7%), while males were only 167 in number (17.3%), bringing the male/female ratio to 1/4.8. In five studies, the demographics were presented only on the initial recruited population [4, 22, 23, 20, and 48].

The aetiology of the coccydynia was reported in 954 patients in all papers except [13, 18, 21, and 30]. The most common cause of coccydynia was direct trauma, recorded in 538 patients from 954 total number of patients with coccydynia included in this study and mentioned the cause of differential causes of it, (56.4%). Not-identifiable causes (idiopathic coccydynia) were present in 232 cases (24.3%), childbirth in 75 females (7.9%), recent rectal or lumbar spinal surgery or epidural injection in 26 (2.7%).

The table includes information regarding operations performed, the use of drains and antibiotics and their follow up

Authors	Operation	Drain	Antibiotic	Follow up
Trollegaard[41]	total coccygectomy	n/s	IV cefuroxime per-operatively	4.9 months
Shao-wen [42]	total coccygectomy	Yes	pre and postoperative for 72 hours	3.3 years (1-6 years)
Khorrami [43]	Key	No	(cephalosporin)30 minutes before and postop. up to 48 hours	25.7 months
Hanley [44]	total coccygectomy	No	Cefazolin peri-operatively and oral Sulfamethoxazole-Trimethoprim (800 mg-160 mg) and Cephalexin (500 mg) for Five days post-operatively	2 years
Jacquot [45]	Described	n/s	two prophylactic antibiotics over a period of 48 h	1 year
Ramieri [46]	Key	n/s	second dose of 2 g Cephazoline. Further antibiotic Levofloxacin was continued for 5 days	33 months (range 24–70)
Haddad [47]	Described	Yes	IV cefuroxime on induction and oral co-amoxiclav continued until the wound drys	24 months
Awwad [48]	Described	n/s	Post-operatively, metronidazole, 500 mg every 8 h, and cefalexin, 500 mg every 6 h, were given orally for 1 week	6 years (2-16 years)
Ogur [49]	Key	n/s	Preop. Antibiotic 2g IV cefazolin	28 months (range 16–48 months)
Wood and Greger [50]	Key	No	Cefazolin are continued for 48 hours postop.	4.8 years (2-9 years)
Wray and Templeton [13]	n/s	n/s	n/s	5.5 years (1-9 years)
Postacchini and Massobrio [7]	Described	n/s	n/s	In 36 pts in 7.8 years
Bayne et al. [5]	Gardner	n/s	No use	7 years
Eng et al. [14]	Partial coccyx	n/s	n/s	1.5 years
Hellberg and Strange-Vognsen [15])	n/s	No	n/s	15 years (1–16 years
Wray et al. [4]	n/s	n/s	n/s	n/s
Grosso and van Dam [16]	n/s	No	Yes: no further data	56 months (26–92)
Zayer [17]	n/s	No	n/s	5 years
Maigne et al. [18]	Described	Yes	n/s	12 and 24 months
Perkins et al. [19]	Key	n/s	n/s	43 months (19–74 months)
Ramsey et al. [20]	Key 7 partial 8 total	n/s	1 pre: 1 post op	14 months (6–24)
Doursounian et al. [21]	Key	Yes	2nd generation/48 h	12
Hodges et al. [22]	n/s	n/s	2nd generation/gentamycin/ Metronidazole	28 (12–70)
Wood and Mehbod [23]	Key	No	2nd generation/48 h	26 months (12–59 months)
Karalezli et al. [24]	Total 11/partial 3	n/s	n/s	30 months (4–48)
Feldbrin et al. [25]	n/s	n/a	n/s	Minimum 12 months
Pennekamp et al. [26]	Sub-periosteal	Yes	1–3 days	7.3 years (2–16)
Balain et al. [27]	n/s	No	n/s	6.75 years (2–16)
Mouhsine et al. [28]	Partial coccyx	No	n/s	2.8 (14 months–6 years)
Capar et al. [29]	Key	n/a	2nd generation and aminoglycoside/72 h	Minimum 9 months
Sehirlioglou et al. [30]	Key	No	2nd generation/48 h	4.1 years (2–8 years)
Cebesoy et al. [31]	n/s	n/s	For 5 days post op	26 months (24–32 months)
Bilgic et al. [12]	Total removal 11 Sub-periost 14	n/a	2nd generation/48 h	20.4 (12–36)
Traub et al. [32]	Described	n/a	1 dose	21.7 months
Arafa et al. [51]	Described	n/s	n/s	4 years

The classification introduced by Postacchini and Massobrio [7] was used in 9 papers [7, 16, 23, 24, 25, 29, 49, 50, and 51].

In total, 153 X-rays were analyzed and according to them, there were 57 type I (37.3%), 48 type II (31.4%), 29 type III (18.9%) and 19 type IV (12.4%) [3, 16, 18, 19, 23, 42, 48]. Grosso and van Dam [16] reported that 4/5 of their patient population was in groups II, III, IV. Some other studies used the subluxation and hypermobile characteristics of the coccyx, presenting some prevalence of the subluxation type over the hypermobile, which was not statistically significant [18, 21, and 28].

There were four (1 prospective) comparative studies assessing the efficiency of injections to surgical treatment (coccygectomy) [4, 20, 22, 23]. Their findings showed that injection with manipulation is effective in providing pain relief, but they also suggested that coccygectomy is a valid surgical option for those in whom the conservative management failed.

Before doing coccygectomy, most of the patients had undergone non-operative treatment modalities for a variable period of time ranging from 3–6 months like in [3, 15, and 22] to 14–15 years [7, 16].

In the majority of the papers, the operation performed was well described. Most of the surgeons prefer a surgical exposure similar to the operation introduced by Key [10].

Preoperatively, the surgeons suggested pharmaceutical constipation and/or a low residual diet mainly to prevent any possible contamination of the wound during the operation.
For the same reason, a fleet enema was used the day before surgery and intravenous administration of second generation cephalosporin was advisable.

The patients had general anaesthesia and were positioned prone with the buttocks separated and secured laterally for facilitating the exposure. A longitudinal incision was performed from the sacrum to the tip of the coccyx (either with knife or electro cautery).