Review Article

AO SPINE

Global Spine Journal
2022, Vol. 12(7) 1611–1623
© The Author(s) 2021
Article reuse guidelines:
sagepub.com/journals-permissions
DOI: 10.1177/21925682211065389
journals.sagepub.com/home/gsj
SAGE

Coccydynia—The Efficacy of Available Treatment Options: A Systematic Review

Gustav Ø. Andersen, BA Med[1], Stefan Milosevic, BA Med[1], Mads M. Jensen, BA Med[1], Mikkel Ø. Andersen, MD[2], Ane Simony, PhD[2], Mikkel M. Rasmussen, PhD[1], and Leah Carreon, MD, MSc[2,3]

Abstract

Study design: Systematic Review

Objective: To evaluate the efficacy of available treatment options for patients with persistent coccydynia through a systematic review.

Methods: Original peer-reviewed publications on treatment for coccydynia were identified using Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines by performing a literature search of relevant databases, from their inception to January 17, 2020, combined with other sources. Data on extracted treatment outcome was pooled based on treatment categories to allow for meta-analysis. All outcomes relevant to the treatment efficacy of coccydynia were extracted. No single measure of outcome was consistently present among the included studies. Numeric Rating Scale, (NRS, 0–10) for pain was used as the primary outcome measure. Studies with treatment outcome on adult patients with chronic primary coccydynia were considered eligible.

Results: A total of 1980 patients across 64 studies were identified: five randomized controlled trials, one experimental study, one quasi-experimental study, 11 prospective observational studies, 45 retrospective studies and unpublished data from the DaneSpine registry. The greatest improvement in pain was achieved by patients who underwent radiofrequency therapy (RFT, mean Visual Analog Scale (VAS) decreased by 5.11 cm). A similar mean improvement was achieved from Extracorporeal Shockwave Therapy (ESWT, 5.06), Coccygectomy (4.86) and Injection (4.22). Although improved, the mean change was less for those who received Ganglion block (2.98), Stretching/Manipulation (2.19) and Conservative/Usual Care (1.69).

Conclusion: This study highlights the progressive nature of treatment for coccydynia, starting with noninvasive methods before considering coccygectomy. Non-surgical management provides pain relief for many patients. Coccygectomy is by far the most thoroughly investigated treatment option and may be beneficial for refractory cases. Future randomized controlled trials should be conducted with an aim to compare the efficacy of interventional therapies amongst each other and to coccygectomy.

Keywords

chronic pain, coccyx, injection, orthopedic

Introduction

Coccydynia is pain located in the coccygeal bone or the surrounding tissues.[1] Coccydynia is a relatively rare condition, occurring more frequently in females and in all ages.[1-3] The anatomy of the os coccygis varies. It consists of a number of rudimentary vertebrae ranging from 3 to 5 and varies in regard to the incidence of segmental fusion. The positioning of the coccyx has been described and classified into 4 types by

[1]Department of Neurosurgery, Aarhus University Hospital, Aarhus, Denmark
[2]Center for Spine Surgery & Research, Middelfart Hospital, Østre Hougvej, Denmark
[3]Norton Leatherman Spine Center, Louisville, KY, USA

Corresponding Author:
Gustav Ø. Andersen, Department of Neurosurgery, Aarhus University Hospital, Palle Juul-Jensens Boulevard 165, Aarhus 8200 Denmark.
Email: gustav.oesterheden.andersen@post.au.dk

Creative Commons Non Commercial No Derivs CC BY-NC-ND: This article is distributed under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs 4.0 License (https://creativecommons.org/licenses/by-nc-nd/4.0/) which permits non-commercial use, reproduction and distribution of the work as published without adaptation or alteration, without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (https://us.sagepub.com/en-us/nam/open-access-at-sage).

Postacchini and Massobrio.[4] Coccydynia is most frequently associated with single-axis traumatic injury, childbirth, obesity, and rapid weight-loss related to gastric by-pass surgery.[5,6] There are several etiologies to the occurrence of secondary coccydynia, such as cancer pain, infection, or iatrogenic.[7] Previous surgery in the area can lead to inflammation, formation of granulation tissue, adhesions, and possibly a change in elasticity of the tissue surrounding the os coccygis, which, over the course of time, can lead to secondary coccydynia. Extracoccygeal disorders may also manifest as coccydynia. Examples of such are pilonidal cysts, perianal abscesses, hemorrhoids, and diseases of the pelvic organs as well as disorders of the lumbosacral spine, sacroiliac joints, piriformis muscle, and the sacrum.[8,9]

Coccydynia presents most frequently in an acute form with mild symptoms, typically resolving with no treatment within weeks to months.[5] When pain does not resolve, treatment is primarily expectant and aimed at symptom management, as pain spontaneously improves in up to 90% of patients receiving conservative treatment.[10] However, for some patients the pain persists and remains refractory to initial conservative treatment.[5] Chronic coccydynia is a condition for which there is limited understanding of the pathology and the effectiveness of different treatments. Patients may experience a marked loss in quality of life and difficulty in performing everyday activities.[5] Sitting is often conspicuously painful in patients with coccydynia, but can be exaggerated with sexual intercourse, with some patients also having difficulty defecating.[3]

There are various treatment options available for symptom relief, including conservative, pharmacological, and surgical treatment. Patients are advised to sit on a U-shaped cushion or a modified wedge-shaped cushion.[10,11] Other options are non-steroidal anti-inflammatory drugs (NSAIDs), massage, stretching, physical therapy,[11,12,13] or interventional treatment, such as steroid injections, radiofrequency treatments (RFT), extracorporeal shockwave therapy (ESWT), and ganglion blocks.[14-19] Surgical intervention, including both partial and complete resection of the coccyx, is typically an option for patients with coccygeal pain refractory to other therapeutic options.[20-23]

Currently there are no official clinical guidelines regarding the treatment of coccydynia. With this systematic review the authors aim to contribute to the development of clinical guidelines for the treatment of coccydynia.

The study objective is to evaluate the efficacy of current available treatments for coccydynia in adults, by systematically reviewing existing original peer-reviewed publications according to the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines.

Materials and Methods

Protocol and Registration

This systematic review was generated following the PRISMA guidelines. A completed copy of the PRISMA checklist is provided (Supplementary Data Content 1, SDC 1).

Search, Information Sources, Eligibility Criteria, and Study Selection

A systematic literature search was conducted on January 17, 2020, in EMBASE.com, PubMed.com, Scopus, and Web of Science bibliographic databases from their inception to the search date. The search was conducted using index-words related to the coccyx and coccydynia. Index-words together with the full search strategy are attached (SDC 2). An experienced librarian, affiliated with the Faculty of Health at Aarhus University, was consulted for guidance in designing the search. A search for published studies and Epubs ahead of print in journals with relevance to spine surgery was also conducted. Finally, reference lists and citations of the included studies were screened in order to identify other relevant papers, and a cohort of non-published data from DaneSpine was included in the review.[24] The inclusion and exclusion criteria were created based on eligibility (SDC 3).

Inclusion Criteria Were

(1) Publications of original peer-reviewed randomized controlled trials, cohort studies, or case-series, available in full text.
(2) Papers in English, Danish, Norwegian, Swedish, Serbian, Croatian, Bosnian, and Spanish.
(3) Studies addressing treatment of patients with coccydynia with any available treatment option.

Exclusion Criteria Were

(1) Animal studies and studies addressing evaluation of technical equipment.
(2) Studies including patients less than 16 years of age.
(3) Studies without treatment outcome (e.g., studies of etiology).
(4) Acute coccydynia or patients with coccydynia with a duration less than 2 months.
(5) Studies solely concerning secondary coccydynia as a complication of another condition (e.g., cancer-derived pain and infectious-derived pain).
(6) Systematic reviews, meta-analyses, opinions, commentaries, and studies involving less than six cases.

Identified articles were screened for duplicates, using both EndNote and Covidence. The screening was done by two authors independently using Covidence, involving a third author in case of disagreement. Articles were initially screened based on title and abstract, followed by a full text screening. In cases where initial screening could not be performed due to a missing abstract, the full text article was obtained.

Risk of Bias in Individual Studies

To assess article quality and bias, two authors independently evaluated all included articles, followed by an attainment of

consensus. Cochrane Risk of Bias Tool was used to assess the presence and extent of bias in Randomized Controlled Trials (RCTs).[25] Included observational studies were scored using Strengthening the Reporting of Observational studies in Epidemiology (STROBE) checklist.[26]

Data Collection Process and Data Items

Data extraction was performed independently in duplicate and compared when completed. During the process of data extraction, all suitable measures of treatment effect were initially extracted due to the largely heterogeneous sample of included studies. The primary outcome measure for evaluating the efficacy of the different treatments was the Visual Analog Scale (VAS) pain scores at last follow-up compared to baseline. Secondary outcome measures consistent throughout the included studies were complications and qualitative measures of outcome, that is, "improved," "no change," "worse." The authors recorded the proportions of these qualitative measures by consensus, considering that some studies used a different terminology, that is, "better," "unchanged," "worse." It was decided to compile the different qualitative measures of outcome into "successful, moderate, or poor outcome" as this was deemed the most representative. In addition, two continuous measures of pain before and after treatment were established as pre- and post-Numeric Rating Scale ranging from 0 to 10. It was decided to interpret several different measures of pain scores, such as Visual Analog Scale (VAS), Pain Analogue Scale (PAS), and Numeric Pain Score (NPS) to the Numeric Rating Scale (NRS). Studies that reported pain using the VAS ranging from 0 to 100 were rescored to a 0 to 10 scale.

Data Analysis

The included studies were divided into groups based on treatment strategies. All analyses were performed using SPSS V26.0 (IBM, Armonk, New York). Data was analyzed using weighted pooled averages. Mean difference in pain scores from baseline to last follow-up were used as measure of an intervention's efficacy.

The current review was conducted in accordance with the protocol and is registered in PROSPERO (PROSPERO ID: CRD42020166379).

Results

2149 references were identified by applying our search string (SDC 2). After removal of duplicates, a total of 930 references were added to title/abstract screening. Sixty three studies were included for data extraction.[1,9-13,15-21,23,27-75] No articles were included through the search in relevant journals, nor through reference and citation screening (see flowchart of literature screening, Figure 1).

The results were collected from 5 randomized controlled trials, 1 experimental study, one quasi-experimental study, 11 prospective studies and 45 retrospective studies. The STROBE scores for the observational studies varied from 3 to 22 points with a mean of 13.8 (Table 1). The 5 RCTs were not included in the main analysis due to incomparability. The study by Mohanty used unique and therefore incomparable outcome measures,[59] and the study by Doursounian only investigated the complications following coccygectomy.[39] Contemporary case-reports, which were identified during the literature search and not included in the main analysis, reported on novel treatments for coccydynia such as oxygen-ozone administration,[76] dorsal root ganglion stimulation,[77] tarsal tunnel block,[78] and platelet-rich plasma injection therapy.[79]

Data on a total of 1980 patients was extracted and grouped into 7 intervention categories **(Research Data file 1)**. The number of patients in each treatment category varied from 78 to 1103. The proportion of female patients ranged from 71 to 90%, and the mean age ranged from 34.8 to 48.2 years across interventions, the total follow-up time varied from 2 weeks to more than 12 months and successful outcome rates ranged from 24 to 85%. Coccygectomy presented with the highest rate of complications at 11%, but data on complications could only be pooled on coccygectomy, RFT, stretching/manipulation, and ganglion block (Table 2).

As post-intervention NRS scores were only available from one study, stretching/manipulation as a treatment was not included in the pooled analysis. If information was unavailable on any field represented in the table, the field was left empty. Values originating from one study solely are in bold font. The results showed the largest difference (5.11 points) in pre- and post-intervention NRS-score for patients treated with RFT. The lowest difference (1.69 points) was identified in patients treated with conservative/usual care (Table 3, Figure 2).

Discussion

This is the most wide-ranging systematic review on treatment modalities for coccydynia and the first systematic review on the topic.

Our main findings suggested overall good outcomes in most of the treatment modalities investigated. The largest patient-reported pain reductions were observed in ESWT, RFT, and coccygectomy. Usual care and stretching/manipulation showed the least reduction in pain. Ganglion blocks showed a modest effect for a shorter period. Coccygectomy, RFT, and ESWT likewise presented with the highest success-rates, respectively. Coccygectomy is by far the most widely represented treatment-modality in terms of eligible studies and patients included for analysis. In terms of complication rates, all treatment modalities showed very little or no complications, except for coccygectomy which showed an overall high complication-rate. Complications were almost exclusively infections, due to the anatomical area of the surgical site, and could be treated with additional antibiotics.

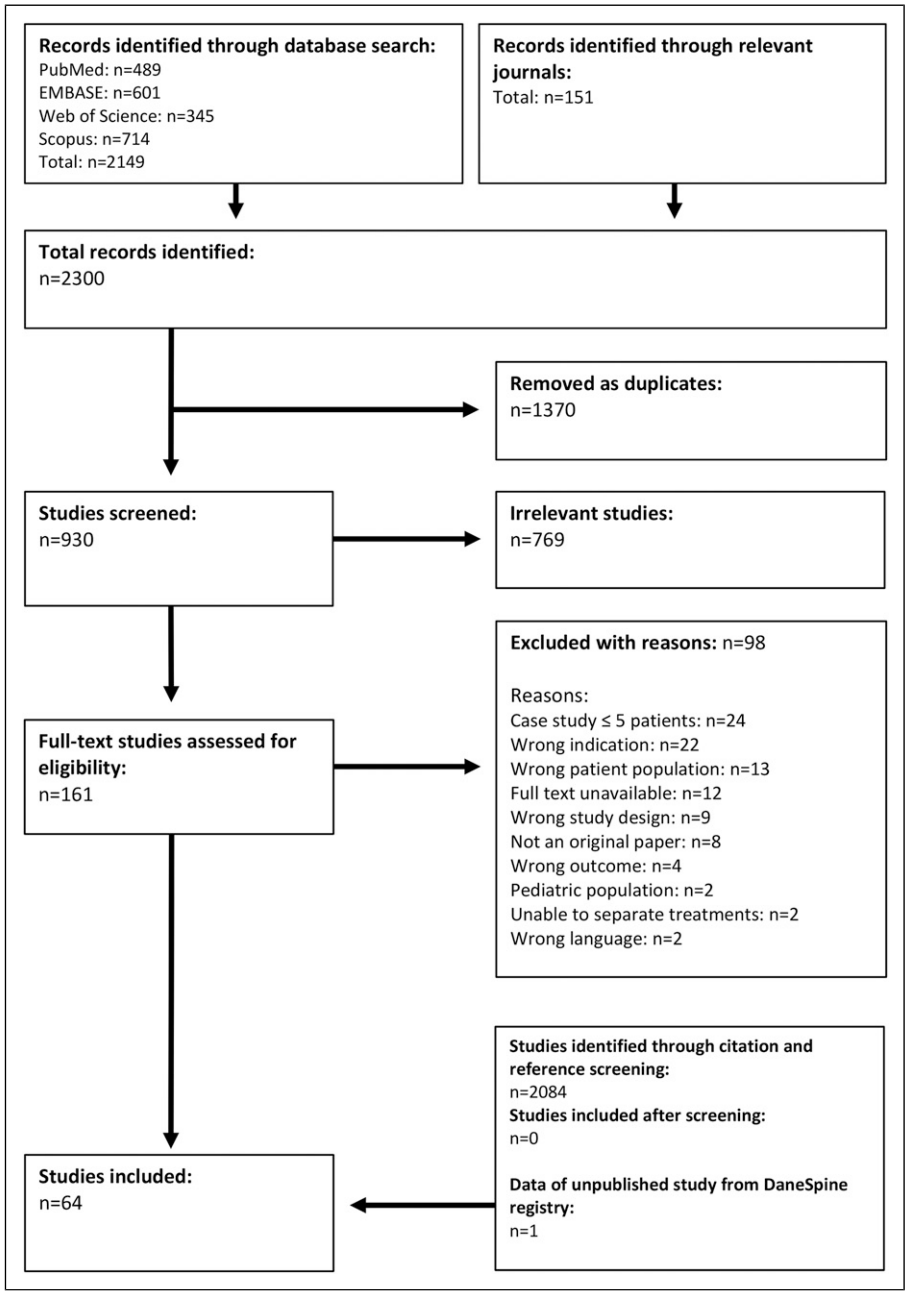

Figure 1. Prisma Flowchart.

The main limitation to this systematic review is the lack of high-quality studies, specifically randomized clinical trials with adequate sample size on the subject. The validity of any analysis is dependent on the quantity and quality of included evidence, which varies widely between the included studies. The limited number of randomized controlled trials, the small sample sizes within the studies and differences in the type of treatments being compared for studies conducted in this field of research proved inadequate for inclusion in the main analysis as no treatment strategies could be pooled. Since the majority of included studies are observational there is inherent bias. Potential bias as a consequence of loss to follow-up and patient selection is present in each study and will inevitably impact the present results. The STROBE score will guide the reader to evaluate the quality of the articles included and the degree of impact this will have on the results. As the investigated interventions were of immense heterogeneity, we considered it too excessive to weight the results of individual studies by their STROBE score. All non-randomized studies of intervention (NRSI) were assessed using STROBE for consistency because of the differences in study design.

Table 1. Studies Included for Analysis in Systematic Review.

Author(s), year	Study Type	Eligible Patients/ Total Patients	Intervention Category	Extracted Outcome	Length of Follow-Up (mos.)	Strengthening the Reporting of Observational Studies in Epidemiology—Score
Abdel-Aal et al.[27]	RCT	60	Conservative/ Usual care	VAS, MMST, ODI	1	-
Adas et al.[15]	Retrospective cohort study	36/41	RFT	VAS, QA	6	18
Alvik and Helsingen.[28]	Retrospective cohort study	13/17	Coccygectomy	QA, Comp	65	7
Antoniadis et al.[1]	Retrospective cohort study	10	Coccygectomy	VAS, QA, Comp	12	17
Awwad et al.[29]	Retrospective cohort study	8/70	Coccygectomy	QA, Comp	72	10
Bayne et al.[9]	Retrospective cohort study	34/48	Coccygectomy	QA, Comp	83	11
Bilgic et al.[30]	Retrospective cohort study	25	Coccygectomy	VAS, QA, Comp	21	17
Bohm.[31]	Retrospective cohort study	14	Rhizotomy	QA, Comp	45	3
Cebesoy et al.[32]	Retrospective cohort study	21	Coccygectomy	VAS, QA	26	14
Chen et al.[16]	Retrospective cohort study	12	RFT	QA, Comp	6	13
Cheng et al.[33]	Retrospective cohort study	31	Coccygectomy	QA, Comp	40	9
Cortiñas Sáenz et al.[34]	Retrospective cohort study	21	Ganglion block	VAS, QA	6	16
Dalbayrak et al, 2014	Retrospective cohort study	32	Coccygectomy, injection	VAS, Comp	N/A	9
Datir and connell.[35]	Retrospective cohort study	8	Ganglion block	VAS, QA	6	10
Demircay et al.[37]	Retrospective cohort study	10	RFT	VNS, EQ-5D	9	16
Doursounian et al.[38]	Retrospective cohort study	61	Coccygectomy	QA, Comp	6	11
Doursounian et al.[39]	Retrospective cohort study	136	Coccygectomy	Comp	N/A	16
Doursounian et al.[40]	Retrospective cohort study	33	Coccygectomy	Paris questionnaire, QA, Comp	>24	16
El Mohsen Arafa et al.[21]	Prospective cohort study	38	Coccygectomy	VAS, QA, Comp	48	15

(continued)

Table 1. (continued)

Author(s), year	Study Type	Eligible Patients/ Total Patients	Intervention Category	Extracted Outcome	Length of Follow-Up (mos.)	Strengthening the Reporting of Observational Studies in Epidemiology—Score
Feldbrin et al.[41]	Retrospective cohort study	7/9	Coccygectomy	QA	>12	13
Finsen.[42]	Retrospective cohort study	11	Injection	QA	64	10
Galhom et al.[13]	Retrospective cohort study	50	Conservative/ Usual care, injection, coccygectomy	QA, Comp	17	14
Gáspár et al.[43]	Retrospective cohort study	32/34	Coccygectomy	VAS, QA	91	15
Gonen Aydin et al.[17]	Retrospective cohort study	34	ESWT	VAS, SF-36, QA	6	15
Gonnade et al.[18]	Prospective cohort study	31	Ganglion block	NRS, ODI, QA	6	18
Gopal and McCrory[44]	Retrospective cohort study	20	RFT	VAS, QA	12	12
Gunduz et al.[45]	Retrospective pilot study	22	Ganglion block	VAS, QA	3 wks	15
Haddad et al.[46]	Retrospective cohort study	14	Coccygectomy	PAS, QA, Comp	80	15
Haghighat and Mashayekhi asl[47]	Quasi-experimental study	10	ESWT	VAS	7	18
Hanley et al.[20]	Prospective cohort study	98	Coccygectomy	VAS, ODI, SF-36, QA, Comp	24	22
Hodges et al.[48]	Retrospective cohort study	11/32	Coccygectomy	VAS, ODI, QA, Comp	>9	15
Karalezli et al.[49]	Retrospective cohort study	14	Coccygectomy	QA, Comp	30	9
Karaman et al.[50]	Retrospective cohort study	8/24	RFT	VAS, QA	9	17
Kerr et al.[51]	Retrospective cohort study	23/26	Coccygectomy	VAS, QA, Comp	37	17
Khan et al.[52]	Prospective cohort study	37	Prolotherapy	VAS, Comp	N/A	11
Kircelli et al.[53]	Retrospective cohort study	20	RFT	VNS, EQ-5D, QA	17	16
Kleimeyer et al.[11]	Retrospective cohort study	88	Coccygectomy, conservative/ Usual care	VAS, EQ-5D, PROMIS, QA, Comp.	58	17
Kulkarni et al.[54]	Retrospective cohort study	10	Coccygectomy	VAS, QA, Comp	21	14

(continued)

Table 1. (continued)

Author(s), year	Study Type	Eligible Patients/ Total Patients	Intervention Category	Extracted Outcome	Length of Follow-Up (mos.)	Strengthening the Reporting of Observational Studies in Epidemiology—Score
Lin et al.[55]	RCT	41	ESWT, interferential current	VAS, ODI, SSS	2	-
Maigne et al.[6]	Prospective cohort study	37	Coccygectomy	QA, Comp	>24	15
Maigne and Chatellier[12]	Prospective pilot study	74	Stretching/Manipulation	VAS, QA	24	21
Maigne et al.[56]	RCT	102	Conservative/Usual care, stretching/Manipulation	VAS, McGill, Paris and Dallas questionnaires, QA	6	-
Margo[57]	Retrospective cohort study	13/318	Coccygectomy	QA	-	7
Marwan et al.[58]	Prospective cohort study	14/17	ESWT	NPS, ODI	4	18
Mohanty and Pattnaik[59]	Experimental study	48	Conservative/Usual care, stretching/Manipulation, Injection	QA	1	16
Mouhsine et al.[10]	Retrospective cohort study	15	Coccygectomy	QA, Comp	32	13
Ogur et al.[60]	Retrospective cohort study	22	Coccygectomy	VAS, QA, Comp	28	15
Perkins et al.[61]	Retrospective cohort study	13	Coccygectomy	NPS, ODI, QA, Comp	43	13
Pyper[62]	Retrospective cohort study	28	Coccygectomy	QA, Comp	42	8
Ramsey et al.[63]	Retrospective cohort study	15/24	Coccygectomy	QA, Comp	14	9
Rubio et al.[64]	Prospective cohort study	6	Ganglion block	QA, Comp	-	12
Sarmast et al.[65]	Prospective cohort study	16	Coccygectomy	VAS, QA, Comp	24	14
Sehirlioglu et al.[66]	Retrospective cohort study	74	Coccygectomy	QA, Comp	49	13
Seker et al.[67]	Retrospective cohort study	44	Injection	VAS, QA	28	16
Sencan et al.[68]	Retrospective cohort study	37	Ganglion block	VAS, QA, Comp	5	18
Sencan et al.[68]	RCT	73	Ganglion block	NRS, Beck test	3	-
Sir and Eksert.[69]	Retrospective cohort study	39	Ganglion block, injection	NPRS, Likert scale, Comp	6	17

(continued)

Table 1. (continued)

Author(s), year	Study Type	Eligible Patients/ Total Patients	Intervention Category	Extracted Outcome	Length of Follow-Up (mos.)	Strengthening the Reporting of Observational Studies in Epidemiology—Score
Sucuoglu et al.[70]	Prospective cohort study	8/128	Injection	VAS	3 wks	18
Traub et al.[71]	Retrospective cohort study	8/10	Coccygectomy	Comp	22	12
Trollegaard et al.[72]	Retrospective cohort study	41	Coccygectomy	QA, Comp	83	16
Wood and Mehbod[73]	Retrospective cohort study	45	Coccygectomy, injectionSSS	QA, Comp	26	14
Wright[74]	Prospective cohort study	12	Chemical ablation	QA	>12	7
Yeganeh et al.[75]	RCT	61	Injection	VAS	2	-

Abbreviations: RCT, Randomized Controlled Trial; VAS, Visual Analog Scale; MMST, Modified Modified Schober Test; ODI, Oswestry Disability Index; RFT, Radiofrequency thermocoagulation; QA, Qualitative Assessment; Comp, Complications; VNS, Visual Numeric Scale; EQ-5D, EuroQoL-5 Domain; ESWT, Extracorporeal Shockwave Therapy; SF-36, Short Form 36; NRS, Numeric Rating Scale; PAS, Pain Analogue Score; VNS, Visual Numeric Scale; PROMIS, Patient-Reported Outcomes Measurement Information System; SSS, Subjective Satisfaction Score; NPS, Numeric Pain Scale; NPRS, Numeric Pain Rating Scale.

Table 2. Summary of Pooled Data Available for Extraction from Included Studies by Type of Intervention.

	Coccygectomy	Conservative/ Usual care	Extracorporeal Shockwave Therapy	Injection	Radiofrequency therapy	Stretching/ Manipulation	Ganglion block	Coccygectomy + DaneSpine
Patients (N)	991	157	78	174	120	125	223	1103
Females (%)	81%	71%	79%	77%	65%	90%	78%	82%
Age, yrs, mean	42.20	46.90	40.71	34.82	48.20	45.18	41.18	42.01
Symptom duration, mos, mean	21.87	**11.00**	27.70	5.47	24.21	**15.00**	16.75	21.87
Length of follow-up, mos, mean	29.79	49.54	5.43	25.29	6.58	**24.00**	5.49	27.71
BMI, kg/m^2, mean	25.98	28.47	25.54		**27.98**	24.24	27.09	26.26
Pain score, 0–10, mean								
Baseline	7.54	6.69	8.13	5.56	7.52	6.24	7.92	7.44
2–6 wks		4.98	3.17	4.18	2.83	**4.05**	3,27	
8–16 wks	**2.70**		2.80	**2.10**	**3.14**		3.97	**2.70**
6 mos	2.96		3.07		2.92		4.94	2.96
≥ 12 mos	2.32	**5.00**		1.34	2.41			2.58
Evaluation of outcome								
Successful (%)	85%	31%	79%	53%	82%	24%	75%	83.4%
Moderate (%)	4%	0%	0%	0%	2%	0%	2%	5.6%
Poor (%)	11%	69%	21%	47%	16%	76%	24%	11%
Complications (%)	11%				8%	0%	1%	12.5%
Evaluation of outcome								
Successful (N)	697/817	32/104	27/34	60/113	79/96	30/124	89/119	775/929
Moderate (N)	32/817	0/104	0/34	0/113	2/96	0/124	2/119	52/929
Poor (N)	88/817	72/104	7/34	53/113	15/96	94/124	28/119	102/929
Complications (N)	104/918				2/26		1/68	128/1026

Abbreviation: BMI, Body Mass Index.

Table 3. Summarized analysis of change in VAS-scores pre- and post- Intervention by type of Intervention. Follow-up score used only if data is from more than one study.

	Baseline	Follow-Up	Difference
Coccygectomy	7.54	2.32	5.22
Coccygectomy + dane	7.44	2.58	4.86
Spine			
Conservative/Usual care	6.69	4.98	1.71
Extracorporeal shockwave therapy	8.13	3.07	5.06
Injection	5.56	1.34	4.22
Radiofrequency therapy	7.52	2.41	5.11
Stretching/Manipulation	6.24		2.19
Ganglion block	7.92	4.94	2.98

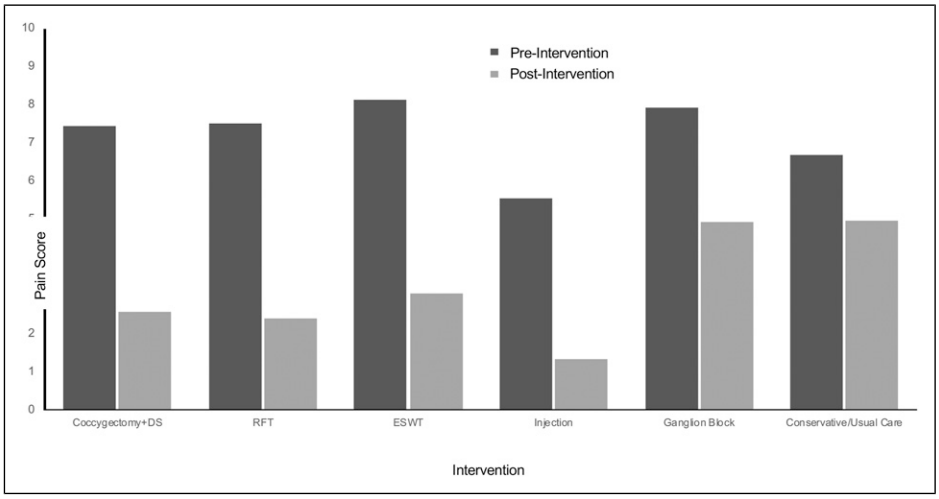

Figure 2. Change in NRS-scores Pre- and Post-Intervention by Type of intervention. Abbreviations: RFT, Radiofrequency thermocoagulation; ESWT, Extracorporeal Shock Wave Therapy.

No comparator to the interventions of interest was noted in the eligibility criteria, due to the sequential nature of the treatment options for coccydynia. Steroid blocks are typically not applied without prior unresponsive attempts at conservative treatment, just as surgical intervention is not performed without prior unresponsive attempts of interventional treatment approach. Furthermore, complete post-operative remission from pain may take months or years after initiation of treatment, why the short-dated follow-up period of some grouped therapies compromises comparability in efficacy across treatments. Due to the study design, the analysis of efficacy does not consider subgroups of patients, for example,, traumatic or idiopathic etiology, which could impact treatment outcome.

As our preliminary research suggested that the amount of studies on treatment options to coccydynia was sparse, our search strategy was constructed without restriction to publication year and without restriction to any types of treatment.

Moreover, we lowered the specificity on outcome measures to avoid exclusion of studies assessing less investigated treatment options and studies not using validated score-systems. This weakens the quality of the overall quantitative comparison in efficacy, as a trade-off to be able to report on efficacy in the largest possible number of patients. In order to include as much data as possible, outcome on eligible patients from mixed patient cohorts was extracted, if data was separable from the remaining study sample. This compromised the availability of patient demographics in some instances.

We also included an unpublished set of consecutively sampled data on the efficacy of coccygectomy with relevance to the review. The unpublished data was included due to the quantitative added value, simultaneous acknowledging the lack of peer-review.

Even though RFT, ESWT, and coccygectomy present with very similar results in the analysis, the validity of our findings regarding RFT and ESWT should be considered in relation to the sparse amount of evidence, whereas coccygectomy is the single most investigated treatment option. Although promising treatments, we consider the basis of the current analysis inadequate for comparing the long-term efficacy of RFT and ESWT to that of coccygectomy.

Despite presenting with the best validated outcome results, surgical treatment should be reserved to a select subset of patients, unresponsive to all available conservative treatment, and interventional treatment options, due to the potential risk of surgical complications. Future randomized controlled trials should be conducted with an aim to compare the efficacy of interventional therapies amongst each other and to coccygectomy.

Conclusion

The results must be interpreted in the context of the patients included for review, which is why noninvasive treatment despite its modest effect should not be discarded as first-line treatment. A sequential nature of treatment stands out all across the literature, and thus, interventional therapy is preferable to invasive treatment as the former often provides pain relief for many patients, but without the evident risk of complications associated with the latter. Coccygectomy is by far the most thoroughly investigated treatment option and may be beneficial for refractory cases to less invasive procedures. High-quality studies in future may obtain the same or completely different results as seen in this systematic review. Future randomized controlled trials should be conducted with an aim to compare the efficacy of interventional therapies amongst each other and to coccygectomy.

Declaration of Conflicting Interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The authors received no financial support for the research, authorship, and/or publication of this article.

ORCID iDs

Gustav Ø. Andersen https://orcid.org/0000-0002-7598-7538
Mads M. Jensen https://orcid.org/0000-0002-5843-6216
Mikkel Ø. Andersen https://orcid.org/0000-0001-8478-8218
Ane Simony https://orcid.org/0000-0002-1788-1161
Leah Carreon https://orcid.org/0000-0002-7685-9036

Ethical Approval

Data use approval was acquired from the Danish Data Protection Agency ref nr: 16/1586. The study was reviewed and approved by the Research Board of the Center for Spine Surgery and Research at Lillebaelt Hospital.

Informed Consent

Subjects provided consent for use of their data at the time patients completed the questionnaires and were enrolled in DaneSpine.

Supplemental material

Supplemental material for this article is available online.

References

1. Antoniadis A, Ulrich NH-B, Senyurt H. Coccygectomy as a surgical option in the treatment of chronic traumatic coccygodynia: A single-center experience and literature review. *Asian Spine J*. 2014;8(6):705-710.
2. Pennekamp PH, Kraft CN, Stutz A, Wallny T, Schmitt O, Diedrich O. Coccygectomy for coccygodynia: Does pathogenesis matter? *J Trauma*. 2005;59(6):1414-1419.
3. Fogel GR, Cunningham PY, Esses SI. Coccygodynia: Evaluation and management. *J Am Acad Orthop Surg*. 2004;12(4):49-54.
4. Postacchini F, Massobrio M. Idiopathic coccygodynia. Analysis of fifty-one operative cases and a radiographic study of the normal coccyx. *J Bone Joint Surg Am*. 1983;65(8):1116-1124.
5. Lirette LS, Chaiban G, Tolba R, Eissa H. Coccydynia: An overview of the anatomy, etiology, and treatment of coccyx pain. *Ochsner J*. 2014;14(1):84-87.
6. Maigne J-Y, Doursounian L, Chatellier G. Causes and mechanisms of common coccydynia—Role of body mass index and coccygeal trauma. *Spine (Phila Pa 1976)*. 2000;25(23):3072-3079.
7. Thiele GH. Coccygodynia; The mechanism of its production and its relationship to anorectal disease. *Am J Surg*. 1950;79(1):110-116.
8. Nathan ST, Fisher BE, Roberts CS. Coccydynia: A review of pathoanatomy, aetiology, treatment and outcome. *J Bone Joint Surg Br*. 2010;92(12):1622-1627.
9. Bayne O, Bateman JE, Cameron HU. The influence of etiology on the results of coccygectomy. *Clin Orthop Relat Res*. 1984;(190):266-272.
10. Mouhsine E, Garofalo R, Chevalley F, et al. Posttraumatic coccygeal instability. *Spine J*. 2006;6(5):544-549.
11. Kleimeyer JP, Wood KB, Lønne G, et al. Surgery for refractory coccygodynia: Operative versus nonoperative treatment. *Spine (Phila Pa 1976)*. 2017;42(16):1214-1219.
12. Maigne JY, Chatellier G. Comparison of three manual coccydynia treatments: A pilot study. *Spine (Phila Pa1976)*. 2001;26(20):E479-E484.
13. Galhom A, Al-Shatouri M, El-Fadl SA. Evaluation and management of chronic coccygodynia: Fluoroscopic guided injection, local injection, conservative therapy and surgery in non-oncological pain. *Egypt J Radiol Nucl Med*. 2015;6(4):1049-1055.
14. Atim A, Ergin A, Bilgic S, Deniz S, Kurt E. Pulsed radiofrequency in the treatment of coccygodynia. *Agri*. 2011;23(1):1-6.
15. Adas C, Ozdemir U, Toman H, Luleci N, Luleci E, Adas H. Transsacrococcygeal approach to ganglion impar: Radiofrequency application for the treatment of chronic intractable coccydynia. *J Pain Res*. 2016;9:1173-1177.

16. Chen Y, Huang-Lionnet JHY, Cohen SP. Radiofrequency Ablation in Coccydynia: A case series and comprehensive, evidence-based review. *Pain Med*. 2017;18(6):1111-1130.
17. Gonen Aydin C, Örsçelik A, Gök MC, Akman YE. The efficacy of extracorporeal shock wave therapy for chronic coccydynia*: Med Princ Pract*;29(5):444-450, 2020.
18. Gonnade N, Mehta N, Khera PS, Kumar D, Rajagopal R, Sharma PK. Ganglion impar block in patients with chronic coccydynia. *Indian J Radiol Imaging*. 2017;27(3):324-328.
19. Sencan S, Edipoglu IS, Ulku Demir FG, Yolcu G, Gunduz OH. Are steroids required in the treatment of ganglion impar blockade in chronic coccydynia? A prospective double-blinded clinical trial. *Korean J Pain*. 2019;32(4):301-306.
20. Hanley EN, Ode G, Jackson III JB, Seymour R. Coccygectomy for patients with chronic coccydynia: A prospective, observational study of 98 patients. *Bone Joint J*. 2016;98-B(4):526-533.
21. El Mohsen Arafa A, El Rady Mahmoud A, Zayan M, Kotb A. Coccygectomy for posttraumatic coccygodynia: A long-term prospective study. *Curr.Orthop.Pract*. 2017;28(5):484-488.
22. Wray C, Easom S, Hoskinson J. Coccydynia. Aetiology and treatment. *J Bone Joint Surg Br*. 1991;73-B(2):335-338.
23. Maigne J-Y, Lagauche D, Doursounian L, et al. Instability of the coccyx in coccydynia. *J Bone Joint Surg Br*. 2000;82(7):1038-1041.
24. DaneSpine. 2019 Årsrapporter. Available from: http://drksdanespine.dk/wm420129.
25. Higgins JPT, Altman DG, Gotzsche PC. The cochrane collaboration's tool for assessing risk of bias in randomised trials. *Bmj*. 2011;343:d5928.
26. von Elm E, Altman DG, Egger M, Pocock SJ, Gøtzsche PC, Vandenbroucke JP. The strengthening the reporting of observational studies in epidemiology (STROBE) statement: Guidelines for reporting observational studies. *J Clin Epidemiol*. 2008;61(4):344-349.
27. Abdel-Aal NM, Elgohary HM, Soliman ES, Waked IS. Effects of kinesiotaping and exercise program on patients with obesity-induced coccydynia: A randomized, double-blinded, sham-controlled clinical trial. *Clin Rehabil*. 2020;34(4):471-479.
28. Alvik E, Helsingen N. [Coccygodynia. A study of 17 patients]. *Tidsskr Nor Laegeforen*. 1976;96(34-36):1799-1800.
29. Awwad WM, Saadeddin M, Alsager JN, AlRashed FM. Coccygodynia review: Coccygectomy case series. *Eur J Orthop Surg Traumatol*. 2017;27(7):961-965.
30. Bilgic S, Kurklu M, Yurttaş Y, Ozkan H, Oguz E, Şehirlioglu A. Coccygectomy with or without periosteal resection. *Int Orthop*. 2010;34(4):537-541.
31. Bohm E. Late results of sacral rhizotomy in coccygodynia. *Acta Chir Scand*. 1962;123:6-8.
32. Cebesoy O, Guclu B, Kose KC, Basarir K, Guner D, Us AK. Coccygectomy for coccygodynia: Do we really have to wait? *Injury*. 2007;38(10):1183-1188.
33. Cheng SW, Chen QY, Lin ZQ, et al. Coccygectomy for stubborn coccydynia. *Chin J Traumatol*. 2011;14(1):25-28.
34. Cortiñas Sáenz M, Iglesias Cerrillo JA, Vázquez Colomo C, Salmerón Vélez G. Infiltration of ganglion impar and caudal in the management of traumatic coccydynia refractory to conventional analgesic treatment. *Revista Espanola de Cirugia Ortopedica y Traumatologia*. 2011;55(5):340-345.
35. Dalbayrak S, Yaman O, Yilmaz T, Yilmaz M. Treatment principles for coccygodynia. *Turk Neurosurg*. 2014;24(4):532-537.
36. Datir A, Connell D. CT-guided injection for ganglion impar blockade: A radiological approach to the management of coccydynia. *Clin Radiol*. 2010;65(1):21-25.
37. Demircay E, Kabatas S, Cansever T, Yilmaz C, Tuncay C, Altinors N. Radiofrequency thermocoagulation of ganglion impar in the management of coccydynia: Preliminary results. *Turk Neurosurg*. 2010;20(3):328-333.
38. Doursounian L, Maigne JY, Faure F, Chatellier G. Coccygectomy for instability of the coccyx. *Int Orthop*. 2004;28(3):176-179.
39. Doursounian L, Maigne J-Y, Cherrier B, Pacanowski J. Prevention of post-coccygectomy infection in a series of 136 coccygectomies. *Int Orthop*. 2011;35(6):877-881.
40. Doursounian L, Maigne J-Y, Jacquot F. Coccygectomy for coccygeal spicule: A study of 33 cases. *Eur Spine J*. 2015;24(5):1102-1108.
41. Feldbrin Z, Singer M, Keynan O, Rzetelny V, Hendel D. Coccygectomy for intractable coccygodynia. *Isr Med Assoc J*. 2005;7(3):160-162.
42. Finsen V. [Corticosteroid injection for coccygodynia]. *Tidsskr Nor Laegeforen*. 2001;121(24):2832-2833.
43. Gáspár L, Kiss L, Vereb G, Csernátony Z. Coccygectomy has a favorable effect on the intensity, manifestation, and characteristics of pain caused by coccygodynia: A retrospective evaluation of 34 patients followed for 3-18 years. *Eur J Orthop Surg Traumatol*. 2009;19(6):403-407.
44. Gopal H, Mc Crory C. Coccygodynia treated by pulsed radio frequency treatment to the Ganglion of Impar: A case series. *J Back Musculoskelet Rehabil*. 2014;27(3):349-354.
45. Gunduz OH, Sencan S, Kenis-Coskun O. Pain relief due to transsacrococcygeal ganglion impar block in chronic coccygodynia: A pilot study. *Pain Med*. 2015;16(7):1278-1281.
46. Haddad B, Prasad V, Khan W, Alam M, Tucker S. Favourable outcomes of coccygectomy for refractory coccygodynia. *Ann R Coll Surg Engl*. 2014;96(2):136-139.
47. Haghighat S, Mashayekhi Asl M. Effects of extracorporeal shock wave therapy on pain in patients with chronic refractory coccydynia: A quasi-experimental study. *Anesth Pain Med*. 2016;6(4):e37428.
48. Hodges SD, Eck JC, Humphreys SC. A treatment and outcomes analysis of patients with coccydynia. *Spine J*. 2004;4(2):138-140.
49. Karalezli K, Iltar S, Irgit K, Karalezli N, Karakoç Y, Aydoğan N. Coccygectomy in the treatment of coccygodynia. *Acta Orthop Belg*. 2004;70(6):583-585.
50. Karaman H, Tüfek A, Kavak GO, Yildirim ZB, Celik F. Would pulsed radiofrequency applied to different anatomical regions have effective results for chronic pain treatment? *JPMA (J Pak Med Assoc)*. 2011;61(9):879-885.
51. Kerr EE, Benson D, Schrot RJ. Coccygectomy coccygectomy for chronic refractory coccygodynia: Clinical case series and literature review. *J Neurosurg Spine*. 2011;14(5):654-663.

52. Khan S, Kumar A, Varshney M, Trikha V, Yadav C. Dextrose prolotherapy for recalcitrant coccygodynia. *J Orthop Surg (Hong Kong)*. 2008;16(1):27-29.
53. Kircelli A, Demirçay D, Özel O, et al. Radiofrequency thermocoagulation of the ganglion impar for coccydynia management: Long-term effects. *Pain Pract*. 2019;19(1):9-15.
54. Kulkarni AG, Tapashetti S, Tambwekar VS. Outcomes of coccygectomy using the "Z" plasty technique of wound closure. *Global Spine J*. 2019;9(8):802-806.
55. Lin S-F, Chen Y-J, Tu H-P, et al. The effects of extracorporeal shock wave therapy in patients with coccydynia: A randomized controlled trial. *PLoS One*. 2015;10(11):e0142475.
56. Maigne J-Y, Chatellier G, Faou M. L, Archambeau M. The treatment of chronic coccydynia with intrarectal manipulation—A randomized controlled study. *Spine*. 2006;31(18):E621-E627.
57. Margo E. The painful terminal spine: A clinicostatistical review of 318 cases of painful terminal spines resulting from fracture, congenital anomalies, ligamentous tear, traumatic or of insidious onset—together with method of treatment and the results therefrom. *South Med J*. 1962;55(5):461-470.
58. Marwan Y, Dahrab B, Esmaeel A, Ibrahim SA, Al-Failakawi J. Extracorporeal shock wave therapy for the treatment of coccydynia: A series of 23 cases. *Eur J Orthop Surg Traumatol*. 2017;27(5):591-598.
59. Mohanty PP, Pattnaik M. Effect of stretching of piriformis and iliopsoas in coccydynia. *J Bodyw Mov Ther*. 2017;21(3):743-746.
60. Ogur HU, Seyfettinoğlu F, Ümit Tuhanioğlu Ü, Cicek H, Zohre S. An evaluation of two different methods of coccygectomy in patients with traumatic coccydynia. *J Pain Res*. 2017;10:881-886.
61. Perkins R, Schofferman J, Reynolds J. Coccygectomy for severe refractory sacrococcygeal joint pain. *J Spinal Disord Tech*. 2003;16(1):100-103.
62. Pyper JB. Excision of the coccyx for coccydynia; A study of the results in twenty-eight cases. *J Bone Joint Surg Br*. 1957;39-b(4):733-737.
63. Ramsey ML, Toohey JS, Neidre A, Stromberg LJ, Roberts DA. Coccygodynia: treatment. *Orthopedics*. 2003;26(4):403-405.
64. Rubio E, Alonso JRS, García-Muñoz M, Meseguer F. Cocygeal transdiscal approach of ganglion impar for the treatment of coccygodynia. *Rev Soc Española Dolor*. 2007;14(2):113-116.
65. Sarmast A, Kirmani A, Bhat A. Coccygectomy for coccygodynia: A single center experience over 5 years. *Asian J Neurosurg*. 2018;13(2):277-282.
66. Sehirlioglu A, Ozturk C, Oguz E, Emre T, Bek D, Altinmakas M. Coccygectomy in the surgical treatment of traumatic coccygodynia. *Injury*. 2007;38(2):182-187.
67. Seker A, Sarikaya IA, Korkmaz O, Yalcin S, Malkoc M, Bulbul AM. Management of persistent coccydynia with transrectal manipulation: results of a combined procedure. *Eur Spine J*. 2018;27(5):1166-1171.
68. Sencan S, Cuce I, Karabiyik O, Demir FU, Ercalik T, Gunduz OH. The influence of coccygeal dynamic patterns on ganglion impar block treatment results in chronic coccygodynia. *Interv Neuroradiol*. 2018;24(5):580-585.
69. Sir E, Eksert S. Comparison of block and pulsed radiofrequency of the ganglion impar in coccygodynia. *Turk J Med Sci*. 2019;49(5):1555-1559.
70. Sucuoğlu H, Özbayrak SS, Uludağ M, Tüzün S. Short-term efficacy of joint and soft tissue injections for musculoskeletal pain: An interventional cohort study. *Agri*. 2016;28(2):79-88.
71. Traub S, Glaser J, Manino B. Coccygectomy for the treatment of therapy-resistant coccygodynia. *J Surg Orthop Adv*. 2009;18(3):147-149.
72. Trollegaard AM, Aarby NS, Hellberg S. Coccygectomy: An effective treatment option for chronic coccydynia—Retrospective results in 41 consecutive patients. *J Bone Joint Surg Series B*. 2010;92-B(2):242-24.
73. Wood KB, Mehbod AA. Operative treatment for coccygodynia. *J Spinal Disord Tech*. 2004;17(6):511-515.
74. Wright BD. Treatment of intractable coccygodynia by transsacral ammonium chloride injection. *Anesth Analg*. 1971;50(4):519-52.
75. Yeganeh A, Taghavi R, Saidifard M, Mahmoudi M, Shahverdi S, Moghtadaei M. Comparing the therapeutic effect of local corticosteroid injections and laser in treating coccyx pains (sacrum). *Biomedical and Pharmacology Journal*. 2015;8:119-123.
76. Bonetti M, Iavarone R, Martinelli F. Coccygodynia treated by oxygen-ozone administration. A case report. *Int J Ozone Therapy*. 2008;7(2):139-142.
77. Giordano NL, van Helmond N, Chapman KB. Coccydynia treated with dorsal root ganglion stimulation. *Case Rep Anesthesiol*. 2018;2018:5832401.
78. Hammodi A. Coccydynia relieved by a tarsal tunnel block: A case series. *J Med Case Rep*. 2019;13(1):339.
79. Montero-Cruz F-R, Aydin SM. Platelet-rich plasma injection therapy for refractory Coccydynia: A case series. *Interventional Pain Management Rep*. 2018;2(5):183-188.