Coccydynia – the efficacy of available treatment options: a systematic review protocol

Mads Moss Jensen (mads.moss.jensen@gmail.com)
Sygehus Lillebalt Middelfart Sygehus https://orcid.org/0000-0002-5843-6216

Gustav Østerheden Andersen
Sygehus Lillebalt Middelfart Sygehus

Stefan Milosevic
Sygehus Lillebalt Middelfart Sygehus

Mikkel Østerheden Andersen
Sygehus Lillebalt Middelfart Sygehus

Leah Carreon
Sygehus Lillebalt Middelfart Sygehus

Ane Simony
Sygehus Lillebalt Middelfart Sygehus

Mikkel Mylius Rasmussen
Aarhus Universitetshospital

Study protocol

Keywords: Coccydynia, Pain, Treatment, Efficacy

Posted Date: April 9th, 2021

DOI: https://doi.org/10.21203/rs.2.22757/v3

Abstract

Background context: Coccydynia is a painful condition which may severely impair quality of life in affected patients. Treatment of coccydynia is a field of interest with limited knowledge. The objective of this study 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.

Methods/design: A systematic literature search will be conducted in EMBASE.com, PubMed.com, Scopus and Web of Science bibliographic databases from their inception to January 17th, 2020, alongside a search for published studies and Epubs ahead of print in journals with relevance to spine surgery. Studies eligible for inclusion are original peer-reviewed papers addressing treatment of chronic coccydynia in adults. The articles will be screened by two authors independently, involving a third author in case of disagreement. Quality assessment and data extraction will be conducted using Covidence software. A meta-analysis will be conducted given that data is suitable.

Discussion: This systematic review and meta-analysis may contribute to the existing knowledge on the efficacy of treatment options for coccydynia.

Systematic review registration: PROSPERO submission-ID: 166379

Background

Coccydynia is pain located in the coccygeal bone or the surrounding tissues[1]. Coccydynia presents most frequently in an acute form with mild symptoms, typically resolving with no treatment within weeks to months[2]. Treatment is primarily expectant and aimed at symptom management, as pain spontaneously improves in up to 90% of patients receiving conservative treatment[3]. However, for some patients the pain persists and remains refractory to initial conservative treatment[2]. Chronic coccydynia is a condition on which there is limited understanding of pathology and the effectiveness of different treatments. These patients may experience a marked loss in quality of life and difficulty in performing everyday activities[2].

The anatomy of the os coccygis varies. It consists of a number of rudimentary vertebrae ranging from three to five and varies in regard to incidence of segmental fusion. The positioning of the coccyx has been described and classified into four types by Postacchini and Massobrio in 1983[4]. It ranges from a caudally pointing coccyx (Type I), over a slightly forward curved coccyx (Type II), to a sharply forward angulated coccyx (Type III). A subluxated coccyx is classified as a Type IV coccyx. A correlation between coccydynia and subluxation or hypermobility of the coccyx has been described, due to instability leading to chronic inflammation and pain[5]. However, Postacchini and Massobrio did not find any statistically significant correlation between the number of coccygeal segments, or incidence of fusion between the segments, and increased risk of coccydynia[4].

Coccydynia is a relatively rare condition, occurring more frequently in females than males and in all ages with a mean age of 40 years[6, 7]. Sitting is often conspicuously painful in patients with coccydynia but can be exaggerated with sexual intercourse, with some patients also having difficulty defecating[7]. Coccydynia most frequently related to single-axis traumatic injury, childbirth, obesity and rapid weight-loss related to gastric by-pass surgery[2, 8].

There are various treatment options available, including conservative, pharmacological and surgical treatment. Patients are advised to sit on a U-shaped cushion, or a modified wedge-shaped cushion, for pain relief[3]. Other options are NSAIDs, massage, stretching or interventional treatment, such as steroid injections and ganglion blocks. 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. Coccygectomy is commonly reported successful with a high rate of pain relief[9].

Currently there are no official clinical guidelines regarding the treatment of coccydynia, which in part is due to an insufficient amount of studies investigating the various treatment options and their outcomes. With this systematic review the authors aim to contribute to the existing knowledge on treatment effect, to guide clinicians when dealing with patients suffering from coccydynia by supplying an overview of existing treatment options and to support any future development of clinical guidelines in 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.

Methods/design

This protocol paper is generated following the PRISMA-P guidelines. A completed copy of the PRISMA-P checklist is provided (Additional file 1).

Eligibility criteria:

Study inclusion will be conducted based on the eligibility criteria outlined in the following Population-Intervention-Comparator-Outcomes-Study design (PICOS) criteria.

Population:

All adult patients ≥ 16 years of age with chronic pain located to the coccyx are considered eligible for inclusion.

Interventions:

Interventions of interest are those of any conservative, interventional or surgical nature described in any eligible paper. The interventions of interest are not further specified, due to the risk of excluding novel

treatments not widely investigated. Papers excluded as case reports will be noted and commented in the discussion section if these concern an intervention of a newer date, not otherwise addressed in the papers included for data analysis.

Comparator:

The authors argue that a comparator is not applicable due to the nature of this study. This is due to the sequential nature of the treatment options for coccydynia. Steroid blocks are typically not applied without prior attempts at conservative treatment, just as surgical intervention is not performed without prior attempts of interventional treatment.

Outcomes:

Outcomes of interest are patient-reported outcome measures, including measures of pain (e.g. VAS-score), quality of life (e.g. EQ-5D), disability (e.g. ODI), work absenteeism, complications to intervention and patient-reported bettering.

Study type:

Studies to be included are any original peer-reviewed literature of randomized control trials, cohort studies or case-series available in full text, both retrospective and prospective, containing more than five patients with coccydynia. Reviews, meta-analyses, opinions and commentaries are excluded, as well as studies involving less than six patients with coccydynia, which will be classified and excluded as case reports.

Language:

Papers in the English, Danish, Norwegian, Swedish, Serbian, Croatian, Bosnian and Spanish language are included.

Exclusion:

Studies that do not meet the eligibility criteria outlined through PICOS are to be excluded, counting animal studies and studies addressing evaluation of technical equipment, studies including patients of less than 16 years of age, studies without treatment outcome (e.g. studies of etiology), studies of acute coccydynia or those with coccydynia reported with a duration less than two months and studies solely concerning secondary coccydynia as a complication to another condition (mimics of coccydynia, e.g. cancer-derived pain and infectious-derived pain).

Secondary coccydynia also includes pain derived from previous surgery in the ano-rectal area. This includes prior coccygectomy, surgery for pilonidal cysts, ano-rectal surgery, as well as any other surgical intervention in the area. This is due to the risk of developing granulation tissue, adhesions, chronic inflammation and possibly a change in elasticity of the tissue surrounding the os coccygis, which, over the course of time, can lead to secondary coccydynia.

Information sources:

A systematic literature search will be conducted in EMBASE.com, PubMed.com, Scopus and Web of Science bibliographic databases from their inception to January 17th 2020, alongside a search for published studies and Epubs ahead of print in journals with relevance to spine surgery, such as Spine, EuroSpine, European Spine Journal, Global Spine Journal, the Spine Journal, International Orthopedics and Journal of Bone and Joint Surgery.

Furthermore, reference-lists and citations of included studies will be searched in order to identify other relevant papers. Finally, a cohort of non-published data from DaneSpine[10] will be included in the review.

The search will be conducted using index-words related to the coccyx and coccydynia. Examples of such are attached (Additional file 2). An experienced librarian, affiliated to the Faculty of Health at Aarhus University, will be consulted for guidance in designing the search.

Screening:

Papers will be screened using both EndNote- and Covidence software for duplicates, following a screening using Covidence software including or excluding papers according to their eligibility outlined through PICOS.

The screening will be done by two authors independently, involving a third author in case of disagreement. Addressing possible inclusion bias, a pilot test of 50 papers will be conducted. Papers will primarily be excluded based on title and abstract only, followed by a full text screening. In cases where initial screening cannot be performed due to the abstract not being available, the full text will be obtained. In order to do full text screening, a thorough search for available full texts will be conducted, should a paper not already be accessible through the reference managing tool.

Risk of bias evaluation:

To assess quality and bias, two authors (MMJ and SM) will independently evaluate all eligible papers, followed by an attainment of consensus, using Covidence software. Cochrane Risk of Bias Tool will be used to assess for the presence and extent of bias in Randomized Clinical Trials (RCTs). As this systematic review will also include observational studies, each study will be scored using Strengthening the Reporting of Observational studies in Epidemiology (STROBE) checklist.

Data extraction:

Data extraction will be performed using the data extraction form within Covidence. Extraction will be done independently in duplicate by two authors (MMJ and SM), including a third author in case of disagreement, and compared when completed.

Data synthesis:

The included studies will be divided into groups based on treatment strategy in order to perform meta-analysis. Data from RCTs will be evaluated separately using RevMan if treatment strategies can be pooled. Data from Observational studies will be analyzed using weighted pool averages, if the amount of

studies included for each treatment strategy is adequate. In case of inadequate data for meta-analysis, or if data is not suited for meta-analysis, a qualitative estimation will be made, by summarizing the study findings. In the case of several eligible studies on a given treatment option, a study will be included in data synthesis corresponding to the applicability of its reported outcome measure.

The primary outcome measure for comparing and evaluating the efficacy of the different treatment options are validated pain-scores, e.g. VAS-score at last follow-up compared to baseline. In studies that report a qualitative measure of outcome, i.e. “improved”, “no change”, “worse”, the authors will record the proportion of patients reporting each outcome for each study by consensus; considering that some studies may use a different terminology, i.e. “better”,” unchanged”, worse. Secondary outcome measures are measures of disability, e.g. Oswestry Disability Index (ODI), quality of life, e.g. EuroQol-5-Domain (EQ-5D), work absenteeism, complications to the treatment and patient-reported effect of treatment.

If any relevant data is missing, the respective authors of the study will be contacted, concerning assessment of the data. If data is inaccessible or insufficient, an estimate will be conducted.

Discussion

The authors predict certain limitations in the process of constructing the systematic review.

The object of research, coccydynia, is a narrow field of investigation. In the preliminary research for this protocol, the authors suspect an uneven data basis for doing meta-analysis in the systematic review. In order to include as many available treatment options as possible, it may be necessary to lower the specificity of outcome measures in order to not exclude studies that report on treatment outcome. In these instances, a qualitative assessment of the study outcome will be made. The rational basis for this is avoiding exclusion of studies assessing less investigated treatment options.

On the matter of data extraction, the authors suspect a diverse process due to an expected range in quality of information between the eligible studies, of which there may only be few, to be included in the review.

Abbreviations

PRISMA-P: Preferred Reporting Items for Systematic Reviews and Meta-Analysis Protocols; VAS: Visual Analogue Scale; NSAIDs: Non-steroid anti-inflammatory drugs.

Declarations

Ethical approval and consent to participate

Not applicable in this protocol paper.

Consent for publication

Not applicable in this protocol paper.

Availability of data and materials

Not applicable in this protocol paper.

Competing interests

The authors declare that they have no competing interests.

Funding

There is no source of funding for this study.

Authors' contributions

GØA, SM and MMJ drafted the protocol, generated the search string for eligible studies and will perform the study selection, assessment of bias, data extraction, analysis and interpretation. LC will supervise and contribute to the statistical analysis. AS generated the dataset to be extracted from DaneSpine[10] database. LC, MØA, AS and MMR have done substantial revision of the protocol. MMR is supervising the protocol development and is the guarantor of the review. All authors read and approved the final manuscript.

Acknowledgements

The authors acknowledge the great help from Anne Vils, an experienced librarian at Aarhus University, Faculty of Health, for her contribution in developing the search strategy.

The authors acknowledge the great help and support from Karen Højmark, Research and Project Nurse at Center for Spine Surgery & Research, Middelfart Hospital.

Additional files

Additional file 1: Preferred Reporting Items for Systematic Reviews and Meta-Analysis Protocols checklist. This file is a completed copy of the PRISMA-Protocols checklist which serves as a tool for the reader to appraise the quality of this protocol. (DOCX)

Additional file 2: Search strategy. This file contains the search string generated for use in EMBASE.com, PubMed.com, Scopus and Web of Science bibliographic databases. (DOCX)

References

1. Antoniadis, A., N.H. Ulrich, and H. Senyurt, *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): p. 705-10.

2. Lirette, L.S., et al., *Coccydynia: An overview of the anatomy, etiology, and treatment of coccyx pain.* Ochsner Journal, 2014. **14**(1): p. 84-87.
3. Mouhsine, E., et al., *Posttraumatic coccygeal instability.* Spine J, 2006. **6**(5): p. 544-9.
4. Postacchini, F. and M. Massobrio, *Idiopathic coccygodynia. Analysis of fifty-one operative cases and a radiographic study of the normal coccyx.* J Bone Joint Surg Am, 1983. **65**(8): p. 1116-24.
5. Patel, R., A. Appannagari, and P.G. Whang, *Coccydynia.* Curr Rev Musculoskelet Med, 2008. **1**(3-4): p. 223-6.
6. Pennekamp, P.H., et al., *Coccygectomy for coccygodynia: does pathogenesis matter?* J Trauma, 2005. **59**(6): p. 1414-9.
7. Fogel, G.R., P.Y. Cunningham 3rd, and S.I. Esses, *Coccygodynia: evaluation and management.* The Journal of the American Academy of Orthopaedic Surgeons, 2004. **12**(1): p. 49-54.
8. Maigne, J.Y., L. Doursounian, and G. Chatellier, *Causes and mechanisms of common coccydynia: role of body mass index and coccygeal trauma.* Spine (Phila Pa 1976), 2000. **25**(23): p. 3072-9.
9. Karadimas, E.J., G. Trypsiannis, and P.V. Giannoudis, *Surgical treatment of coccygodynia: an analytic review of the literature.* Eur Spine J, 2011. **20**(5): p. 698-705.
10. DaneSpine. *Årsrapporter*. 2019 28-06-2019 22-01-2020]; Available from: http://drksdanespine.dk/wm420129.

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- AdditionalfileIPRISMAPchecklist.docx
- Additionalfile2searchstrategy.docx
- LettertoEditor30Mar2021.docx
- SRprotocolhighlightedchanges30Mar2021.docx