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

Protocol

Keywords: Coccydynia, Pain, Treatment, Efficacy

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Abstract

Background: 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

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

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, The Spine, EuroSpine, Global Spine, the Spine Journal, International Orthopedics and Journal of Bone and Joint.

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.

Articles will be screened using both EndNote- and Covidence software for duplicates, following a screening using Covidence software including or excluding articles according to the inclusion- and exclusion criteria. The inclusion and exclusion criteria are created according to the eligibility criteria (Additional file 3).

The inclusion criteria are:

- Publications of original papers available in full text.
- Papers in English, Danish, Norwegian, Swedish, Serbian, Croatian, Bosnian and Spanish language.
- Studies addressing treatment of patients with coccydynia with any available treatment option.

The exclusion criteria are:

- 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).
- Acute coccydynia or patients with coccydynia reported with a duration less than two
- Studies solely concerning secondary coccydynia as a complication to another condition (mimics of coccydynia, e.g. cancer-derived pain and infectious-derived pain).
- Systematic reviews, opinions and commentaries.

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 articles will be conducted. Articles 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 article will be obtained.

To assess article quality and bias, two authors will independently evaluate all eligible articles, followed by an attainment of consensus, using Covidence software. Data extraction will likewise be performed using Covidence, independently in duplicate, and compared when completed.

The included articles will be divided into groups based on treatment strategy, in order to perform meta-analysis. The meta-analysis will be performed using STATA software, in each separate group, 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. 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 as 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 article.

Consent for publication

Not applicable in this protocol article.

Availability of data and materials

Not applicable in this protocol article.

Competing interests

The authors declare that they have no competing interests.

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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.

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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)

Additional file 3: Study eligibility criteria. This file contains information on population, intervention, comparison, outcome, study design, timeframe, language and publication status. (DOCX)

Supplementary Files

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- Additionalfile1PRISMAPchecklist.docx
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