

Clinical Trial



COCCYGEOPLASTY: AN EXPLORATION OF A NOVEL APPROACH FOR TREATING RESISTANT COCCYDYNIA IN PATIENTS WITH COCCYGEAL HYPERMOBILITY

L. Manfrè¹, I. Gil², T. Baptista³, P. Calvão Pires^{4,5}, A.E. de Vivo¹, S. Masala⁶, R.V. Chandra^{7,8}, G. D'Anna⁹, J.A. Hirsch¹⁰, M. Frigerio¹¹ and M. Bonetti¹¹

¹Department of Radiology, IOM Mediterranean Oncology Institute, Viagrande, Sicily, Italy;

²Department of Neuroradiology, Centro Hospitalar do Algarve EPE, Faro, Portugal;

³Department of Neuroradiology, Oxford University Hospitals NHS Trust, Oxford, UK;

⁴Department of Neuroradiology, Centro Hospitalar de Lisboa Central, Lisboa, Portugal;

⁵Department of Neuroradiology, Centro Hospitalar de Lisboa Ocidental EPE, Lisboa, Portugal;

⁶Department of Diagnosis Imaging Interventional Radiology and Clinical Pathology, Ospedale San Giovanni Battista, Roma, Italy;

⁷Department of Interventional Neuroradiology, Monash Health, Clayton, Victoria, Australia;

⁸Department of Image, Monash University Faculty of Medicine Nursing and Health Sciences, Clayton, Victoria, Australia; ⁹Department of Radiology, IRCCS Istituto Clinico Humanitas, Rozzano, Italy;

¹⁰NeuroEndovascular Program, Massachusetts General Hospital, Boston, Massachusetts, USA;

¹¹Department of Neuroradiology, Istituto Clinico Città di Brescia, Brescia, Italy.

Correspondence to: Dr Luigi Manfrè, MD Minimal Invasive Spine Therapy Department, Mediterranean Institute for Oncology, Corso Italia 10, 95129 Catania, Italy e-mail: lmanfre@me.com

ABSTRACT

Coccydynia can be attributed to various factors, including fractures, subluxations, and hypermobility within the sacrococcygeal area. Current treatment options often fall short in effectiveness. Coccygeoplasty (CP) represents a relatively recent, minimally invasive approach that aims to tackle this challenging clinical issue. The aim of this study is to evaluate clinical outcomes immediately following the procedure and at 3- and 12-month follow-ups for patients suffering from coccydynia linked to coccygeal hypermobility and subluxation. Furthermore, we seek to assess any correlations between imaging results and clinical outcomes at the follow-up intervals. A prospectively maintained database was used to retrospectively assess all patients who received CP for chronic coccydynia from January 2005 until December 2023. Each participant exhibited painful hypermobility (greater than 25°) with anterior flexion verified through radiological assessments. Alternative coccydynia causes were ruled out using CT and MRI imaging techniques. Procedures were conducted under local anesthesia with a combination of fluoroscopic and CT guidance. Clinical assessments were performed at 3- and 12-months post-treatment utilizing the Visual Analogue Scale (VAS). A total of 19 patients underwent treatment at a single center. There were no complications linked to the procedures. At both the 3- and 12-months post-treatment, 75% of patients reported substantial reduction in VAS scores compared to baseline, with average reductions of 3.5 and 4.9, respectively. No instances of pain recurrence were noted at the 12-month follow-up,

Received: 23 March 2024	Copyright © by LAB srl 2024
Accepted: 18 April 2024	This publication and/or article is for individual use only and may not be
	further reproduced without written permission from the copyright
	holder. Unauthorized reproduction may result in financial and other
	penalties. Disclosure: All authors report no conflicts of interest relevant
	to this article.

L. Manfrè et al.

although one patient did not experience any pain alleviation. Post-treatment CT scans confirmed the fusion of sacrococcygeal segments in 14 patients, yet no significant correlation was identified between the imaging outcomes and clinical results (p=0.1). Patients suffering from chronic coccygeal pain due to subluxation and hypermobility exhibited positive clinical outcomes following CP, as evidenced at both the 3- and 12-month evaluations. Additional research is warranted to validate this technique further and identify factors that predict treatment success. Coccygeoplasty may serve as a viable alternative to coccygectomy.

KEYWORDS: coccydynia, coccyx, pain, fractures, subluxations, hypermobility

INTRODUCTION

The coccyx, often described as an inverted triangular structure at the base of the spine, typically comprises three to five fused segments (1). The joint connecting the sacrum and coccyx features an interposed fibrocartilage and synovial membrane that permits enhanced mobility under certain conditions, such as during pregnancy (2). Since its initial description by Simpson in 1859, coccydynia has been characterized as pain localized to the coccyx area without significant radiating discomfort. Pain that endures for more than two months is classified as chronic (3). Women between the ages of 30 and 40 are the most prevalent demographic affected, thought to be due to their anatomical configuration making the coccyx more vulnerable to injuries (4-6).

The spectrum of potential causes for coccydynia includes trauma, especially falls while seated, as well as repetitively induced microtrauma from activities like cycling, motorcycling, or horseback riding (6, 7).

Management options for coccydynia encompass conservative approaches as well as traditional surgical interventions. Conservative methods for alleviating pain include physiotherapy techniques, such as pelvic relaxation massage using supportive sitting aids like a donut pillow, non-steroidal anti-inflammatory medications, and warm baths. Additionally, techniques such as intrarectal manipulation of the coccyx and fluoroscopically guided steroid injections may be utilized. Although not employed in the patients discussed in this case series, more invasive conservative treatments are available, including radiofrequency ablation of the coccygeal discs and Walther's ganglion. In instances of chronic pain, a surgical procedure to remove the coccyx, referred to as coccygectomy, may be indicated (5-17).

Coccygeoplasty (CP), a technique inspired by vertebral augmentation methods, has recently emerged as a therapeutic option. This involved the percutaneous injection of polymethylmethacrylate (PMMA) cement into the sacrococcygeal segments. Although still rare, the limited literature available consists of reports discussing its application. The procedure aims to provide stability in cases where hypermobility or subluxation contributes to coccygeal pain (18-21).

The aim of this study is to present clinical outcomes at the procedure's initiation and follow-up periods of 3 and 12 months for individuals diagnosed with coccydynia resulting from subluxation and coccygeal hypermobility. It also aims to evaluate any associations between the imaging findings and clinical outcomes observed during the follow-ups.

MATERIALS AND METHODS

Eligible patients who underwent coccygeoplasty at a single center from January 2005 to December 2023 were selected based on a meticulously maintained database. This study included adult individuals over 18 years old with chronic painful coccygeal subluxation and hypermobility, defined as a greater than 25° difference between standing and seated X-ray imaging. Patients experienced pain localized to the coccyx region, which was resistant to conservative treatments for at least six months and led to significant functional impairment (22). All participants exhibited hypermobility and subluxation, which was evident on CT or dynamic radiographs of the sacrococcygeal region taken in both seated (painful) and standing positions. Subluxation and hypermobility of the coccyx were characterized by flexion exceeding 25° and luxation indicated by more than 25% displacement. Additionally, an MRI was conducted for surgical planning and to exclude other conditions in the sacrococcygeal region that could mimic coccygeal symptoms (Fig. 1).



Fig. 1. MR before treatment documenting fracture-dislocation of the body of the second coccygeal vertebra (arrow).

MRI scans were performed on a 1.5T machine, acquiring sagittal and axial T1SE and T2STIR images without contrast. Pre-treatment spiral CT scans were obtained at 1 mm intervals, with both 2D and 3D sagittal and coronal reconstructions, to facilitate procedure preparation. Post-treatment scans were conducted under the same parameters to evaluate outcomes, including the degree of fusion, cement filling of sacrococcygeal segments, and any cement leakage. Clinically, outcomes were assessed using the Visual Analogue Scale (VAS) prior to the intervention. This clinical study adhered to European Union privacy regulations and received approval from the hospital's ethics board.

Coccygeoplasty Procedure

All participants provided informed consent prior to the procedure. Patients were positioned prone on the CT table. Initially, a spiral CT study was performed to determine the appropriate angulation for the working needle. Although pre-procedure dynamic studies indicated hypermobility in the target area, once positioned prone, no patient demonstrated angulation exceeding 25°. A single Jamshidi-type needle was inserted along the midline, from the S4 level through to the coccyx.

The procedure was conducted under local anesthesia using lidocaine 2% as the sole agent, with no sedation or general anesthesia involved. Continuous monitoring of blood pressure, pulse oximetry, and heart rate was performed throughout the intervention. Antibiotics (1 gram of cefazolin) were administered approximately one hour before the procedure and continued for two days at 12-hour intervals. The procedure took place in a hybrid operating room utilizing a C-arm and CT combination for monitoring (20, 23, 24). A 13-gauge beveled trocar and high-density cement were used for all patients. The needle was introduced through the mid-axis of the sacrum towards the coccyx, and PMMA was injected while gently withdrawing the needle to fill both the coccyx and the caudal sacrum. The foramina was avoided due to the medial placement of the needle. Cement injection was carried out under C-arm fluoroscopy. A follow-up CT scan with 2D reconstruction was obtained immediately after the procedure (Fig. 2-5).



Fig. 2. *CT* control: coccygeoplasty was performed with one needle along the midline, from the level of S4, passing through the coccyx.



Fig. 3. CT control axial view (arrow).



Fig. 4. Spiral CT, sagittal reconstruction of control of the distribution of medical cement (arrows).



Fig. 5. CT spiral, coronal reconstruction to control the distribution of medical cement (arrows).

According to institutional protocol, patients were monitored in the hospital for 48 hours post-procedure and were permitted to ambulate four hours following the intervention. No complications were observed during this period.

Follow-Up

Patients were discharged two days following their procedure, with no subsequent antibiotic treatments required. Clinical outcomes were documented upon discharge, with follow-up evaluations scheduled for 3 and 12 months afterward, assessing patient satisfaction alongside VAS scores. A clinical success was defined by a decrease of at least 2 cm in the VAS scores.

Statistical analysis

Descriptive analyses and comparisons between final imaging findings and clinical outcomes at the 3- and 12month follow-ups employed SPSS software, specifically utilizing the chi-square test for correlation analysis.

RESULTS

Baseline characteristics

The cohort comprised 16 women and 3 men, averaging 47 years of age. Preoperative MRI scans revealed no alternative pathological findings. Each patient had previously engaged in conservative management strategies, including non-steroidal anti-inflammatory medications, with no clinical improvement. Additionally, out of the 19 patients, 13 had undergone prior steroid injections targeting the pudendal plexus, while all had received intrarectal coccygeal manipulation treatments. None had undergone radiofrequency interventions.

Technical findings

Of the patients, 14 achieved complete fusion of sacrococcygeal segments post-procedure, while 5 exhibited incomplete fusion. PMMA had filled all sacrococcygeal segments, and there were instances of cement leakage into surrounding areas in 3 individuals, but none into the central spinal canal; these leakages were deemed asymptomatic.

Clinical outcomes

Patients experienced a marked decrease in VAS scores at the 3-month (mean score reduced from 7.5 to 4.0) and 12-month follow-ups (mean score reduced to 2.6). The average changes in VAS scores were -3.5 and -4.9, respectively. Of the 19 patients, 75% experienced clinical success with a reduction greater than 2. At the one-year mark, 4 patients reported varying levels of coccygeal discomfort, with one patient experiencing no pain relief and three others achieving minimal changes below the predetermined threshold at either follow-up.

Analysis identified no meaningful correlation between technical radiographic outcomes and clinical results (p=0.1).

DISCUSSION

This preliminary study suggests that coccygeoplasty serves as a feasible treatment modality for individuals suffering from refractory coccydynia due to subluxation and hypermobility. The absence of complications further underscores the procedure's safety. In this patient group, there was a significant majority who reported notable pain alleviation, indicating the procedure's potential effectiveness. With additional research, coccygeoplasty has the potential to become a credible alternative to coccygectomy.

The proper selection of patients is essential for successful outcomes in coccydynia cases. Here, the focus was on individuals enduring chronic pain for over six months, presenting clear evidence of subluxation and hypermobility through dynamic imaging. Historical data reflects that a sizeable percentage of those with coccydynia exhibit signs of subluxation or hypermobility as contributing factors.

The role of MRI in this context remains somewhat ambiguous; it primarily assists in excluding other potential pathologies rather than providing definitive insights into typical coccygeal conditions (25-28).

Percutaneous vertebroplasty techniques, originally introduced in the late 1980s (28), are currently regarded as the standard practice for managing certain types of vertebral compression fractures. Drawing parallels between these procedures, CP aims to provide stabilization in cases of hypermobility or subluxation, which in turn may alleviate pain (29).

L. Manfrè et al.

The methodology described in this study diverges from prior reports on coccygeoplasty, utilizing a single-needle approach that targets the sacrococcygeal axis directly. No complications were observed from this method, further validating its safety in the absence of critical structures at the procedural site. CT imaging played a crucial role in securing accurate needle placement (30-32).

In summary, the lack of symptomatic complications from the treatment suggests coccygeoplasty's suitability for well-selected patients. While some individuals did not achieve the desired level of pain relief, they did not exhibit worsening pain, affirming that the technique warrants consideration for those unresponsive to conservative treatments.

Coccygectomy, typically a last-resort measure, carries inherent risks and complications, including prolonged pain, infections, and rare serious adverse effects. Thus, less invasive alternatives such as coccygeoplasty should be considered prior to resorting to surgical interventions (33).

The study's limitations include its retrospective design, which may introduce biases. Although utilizing the VAS as a measure of clinical improvement is widely recognized, additional validation for this specific context may be needed. The fact that only a single, experienced practitioner conducted all procedures may raise questions about wider applicability. Furthermore, the lengthy recruitment period of over 18 years for just 19 patients highlights challenges in organizing such studies.

Despite these considerations, we advocate for the broader application of this approach, which could pave the way for larger-scale studies in the future.

CONCLUSIONS

Findings from this preliminary experience suggest that coccygeoplasty is a promising treatment for patients suffering from refractory coccydynia due to subluxation and hypermobility. Most patients reported meaningful pain relief following the procedure. Further investigations are necessary to substantiate this technique and identify factors that may influence treatment outcomes. Coccygeoplasty should be explored as a potential preference when considering coccygectomy for patients experiencing this condition.

REFERENCES

- 1. 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.
- Berglundh T, Armitage G, Araujo MG, et al. Peri-implant diseases and conditions: Consensus report of workgroup 4 of the 2017 World Workshop on the Classification of Periodontal and Peri-Implant Diseases and Conditions. J Clin Periodontol. 2018;45 Suppl 20(S286-S291. doi:https://doi.org/10.1111/jcpe.12957
- Maigne JY, 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. doi:https://doi.org/10.1097/00007632-200012010-00015
- 4. Hellberg S, Strange-Vognsen HH. Coccygodynia treated by resection of the coccyx. *Acta Orthop Scand*. 1990;61(5):463-465. doi:https://doi.org/10.3109/17453679008993564
- 5. De Andres J, Chaves S. Coccygodynia: a proposal for an algorithm for treatment. *J Pain.* 2003;4(5):257-266. doi:https://doi.org/10.1016/s1526-5900(03)00620-5
- 6. Frantz FW. Indications and guidelines for pectus excavatum repair. *Curr Opin Pediatr.* 2011;23(4):486-491. doi:https://doi.org/10.1097/MOP.0b013e32834881c4
- Maigne JY, Rusakiewicz F, Diouf M. Postpartum coccydynia: a case series study of 57 women. *Eur J Phys Rehabil* Med. 2012;48(3):387-392.
- 8. Maigne JY, Chatellier G, Faou ML, Archambeau M. The treatment of chronic coccydynia with intrarectal manipulation: a randomized controlled study. *Spine (Phila Pa 1976)*. 2006;31(18):E621-627. doi:https://doi.org/10.1097/01.brs.0000231895.72380.64
- 9. Mitra R, Cheung L, Perry P. Efficacy of fluoroscopically guided steroid injections in the management of coccydynia. *Pain Physician*. 2007;10(6):775-778.
- Kwon HD, Schrot RJ, Kerr EE, Kim KD. Coccygodynia and coccygectomy. *Korean J Spine*. 2012;9(4):326-333. doi:https://doi.org/10.14245/kjs.2012.9.4.326
- 11. 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. doi:https://doi.org/10.1097/01.ta.0000195878.50928.3c
- 12. 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. doi:https://doi.org/10.1016/j.injury.2007.01.022

L. Manfrè et al.

- Sorensen ST, Kirkegaard AO, Carreon L, Rousing R, Andersen MO. Vertebroplasty or kyphoplasty as palliative treatment for cancer-related vertebral compression fractures: a systematic review. *Spine J.* 2019;19(6):1067-1075. doi:https://doi.org/10.1016/j.spinee.2019.02.012
- 14. Kam NM, Maingard J, Kok HK, et al. Combined Vertebral Augmentation and Radiofrequency Ablation in the Management of Spinal Metastases: an Update. *Curr Treat Options Oncol.* 2017;18(12):74. doi:https://doi.org/10.1007/s11864-017-0516-7
- 15. White WD, Avery M, Jonely H, Mansfield JT, Sayal PK, Desai MJ. The interdisciplinary management of coccydynia: A narrative review. *PM R*. 2022;14(9):1143-1154. doi:https://doi.org/10.1002/pmrj.12683
- Daily D, Bridges J, Mo WB, Mo AZ, Massey PA, Zhang AS. Coccydynia: A Review of Anatomy, Causes, Diagnosis, and Treatment. JBJS Rev. 2024;12(5):doi:https://doi.org/e24.0000710.2106/JBJS.RVW.24.00007
- 17. Sciubba DM, Pennington Z, Colman MW, et al. Spinal metastases 2021: a review of the current state of the art and future directions. *Spine J.* 2021;21(9):1414-1429. doi:https://doi.org/10.1016/j.spinee.2021.04.012
- Dean LM, Syed MI, Jan SA, et al. Coccygeoplasty: treatment for fractures of the coccyx. J Vasc Interv Radiol. 2006;17(5):909-912. doi:https://doi.org/10.1097/01.RVI.0000217953.74013.87
- Akar E, Koban O, Ogrenci A, Yilmaz M, Dalbayrak S. Polymethylmetacrylate Cement Augmentation of the Coccyx (Coccygeoplasty) for Fracture: A Case Report. *Balkan Med J.* 2020;37(6):348-350. doi:https://doi.org/10.4274/balkanmedj.galenos.2020.2020.4.68
- Zygourakis CC, DiGiorgio AM, Crutcher CL, 2nd, et al. The Safety and Efficacy of CT-Guided, Fluoroscopy-Free Vertebroplasty in Adult Spinal Deformity Surgery. *World Neurosurg.* 2018;116(e944-e950. doi:https://doi.org/10.1016/j.wneu.2018.05.139
- 21. De Leacy R, Chandra RV, Barr JD, et al. The evidentiary basis of vertebral augmentation: a 2019 update. J Neurointerv Surg. 2020;12(5):442-447. doi:https://doi.org/10.1136/neurintsurg-2019-015026
- 22. 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. doi:https://doi.org/10.5812/aapm.37428
- 23. Caudana R, Renzi Brivio L, Ventura L, Aitini E, Rozzanigo U, Barai G. CT-guided percutaneous vertebroplasty: personal experience in the treatment of osteoporotic fractures and dorsolumbar metastases. *Radiol Med.* 2008;113(1):114-133. doi:https://doi.org/10.1007/s11547-008-0230-1
- 24. Manfre L. CT-Guided Posterior Lumbar Interbody Fusion and Distraction. A Case Report. *Neuroradiol J.* 2011;24(6):919-923. doi:https://doi.org/10.1177/197140091102400616
- 25. Mouhsine E, Garofalo R, Chevalley F, et al. Posttraumatic coccygeal instability. *Spine J.* 2006;6(5):544-549. doi:https://doi.org/10.1016/j.spinee.2005.12.004
- 26. Fogel GR, Cunningham PY, 3rd, Esses SI. Coccygodynia: evaluation and management. *J Am Acad Orthop Surg.* 2004;12(1):49-54. doi:https://doi.org/10.5435/00124635-200401000-00007
- 27. Woon JT, Maigne JY, Perumal V, Stringer MD. Magnetic resonance imaging morphology and morphometry of the coccyx in coccydynia. *Spine (Phila Pa 1976)*. 2013;38(23):E1437-1445. doi:https://doi.org/10.1097/BRS.0b013e3182a45e07
- 28. Galibert P, Deramond H, Rosat P, Le Gars D. [Preliminary note on the treatment of vertebral angioma by percutaneous acrylic vertebroplasty]. *Neurochirurgie*. 1987;33(2):166-168.
- 29. Tian QH, Sun XQ, Lu YY, et al. Percutaneous Vertebroplasty for Palliative Treatment of Painful Osteoblastic Spinal Metastases: A Single-Center Experience. J Vasc Interv Radiol. 2016;27(9):1420-1424. doi:https://doi.org/10.1016/j.jvir.2016.04.033
- Pitton MB, Herber S, Koch U, Oberholzer K, Drees P, Duber C. CT-guided vertebroplasty: analysis of technical results, extraosseous cement leakages, and complications in 500 procedures. *Eur Radiol.* 2008;18(11):2568-2578. doi:https://doi.org/10.1007/s00330-008-1020-z
- 31. Kerr EE, Benson D, Schrot RJ. Coccygectomy for chronic refractory coccygodynia: clinical case series and literature review. *J Neurosurg Spine*. 2011;14(5):654-663. doi:https://doi.org/10.3171/2010.12.SPINE10262
- 32. Afzal S, Dhar S, Vasavada NB, Akbar S. Percutaneous vertebroplasty for osteoporotic fractures. *Pain Physician*. 2007;10(4):559-563.
- 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. doi:https://doi.org/10.1016/j.injury.2006.09.013