osteopoikilosis and fracture healing: the bone quantity–quality paradox—a case report and systematic review with synthesis without meta-analysis

Orthopedic Reviews

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eMethods 1: PRISMA/SWiM checklist

Section and Topic	Item #	Checklist item	Location where item is reported
TITLE			
Title	1	Identify the report as a systematic review.	Title
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	Abstract
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	Introduction
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	Introduction
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	eTable1
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	Search strategy
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	eMethods 4
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	Eligibility criteria and eTable 2&3
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	Data extraction
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	Data extraction
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	Table 2
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	Quality assessment and eMethods 5

Section and Topic	Item #	Checklist item	Location where item is reported
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	Data synthesis
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	Data synthesis
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	Data synthesis and eMethods 2
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	Data synthesis
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	Data synthesis
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	eMethods 6
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	eMethods 6
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	Data synthesis
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	Data synthesis
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	Figure 7
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	eTable 3
Study characteristics	17	Cite each included study and present its characteristics.	Results and Table 2
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	eTable 4
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	Results
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	Results
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	Not applicable; no

Section and Topic	Item #	Checklist item	Location where item is reported
			quantitative synthesis or meta-analysis was performed due to the descriptive nature of the available evidence and absence of comparable effect estimates.
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	eMethods 6 and Result
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	Not applicable; no quantitative synthesis or meta-analysis was performed due to the descriptive nature of the available evidence and absence of comparable effect estimates.
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	eTables 2&3
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	Discussion

Section and Topic	Item #	Checklist item	Location where item is reported
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	Discussion
	23b	Discuss any limitations of the evidence included in the review.	Limitations
	23c	Discuss any limitations of the review processes used.	Discussion
	23d	Discuss implications of the results for practice, policy, and future research.	Discussion
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	Methods
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	Methods
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	eMethods 3
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	Declarations
Competing interests	26	Declare any competing interests of review authors.	Declarations
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	Supplementary Materials: eMethods 1–6; eTables 1–4 , Additional data available upon reasonable request

eMethods 2: Additional SWiM reporting items

Checklist for Synthesis Without Meta-analysis (Campbell *et al.*, 2020) [1].

Campbell M, McKenzie JE, Sowden A, Katikireddi SV, Brennan SE, Ellis S, Hartmann-Boyce J, Ryan R, Shepperd S, Thomas J, Welch V, Thomson H. Synthesis without meta-analysis (SWiM) in systematic reviews: reporting guideline BMJ 2020;368:l6890 http://dx.doi.org/10.1136/bmj.l6890

SWiM reporting item	Item description	Page in manuscript where item is reported	Other*
Methods			
1 Grouping studies for synthesis	1a) Provide a description of, and rationale for, the groups used in the synthesis (e.g., groupings of populations, interventions, outcomes, study design)	Data synthesis & eMethods 6	
	1b) Detail and provide rationale for any changes made subsequent to the protocol in the groups used in the synthesis	eMethods 3	
2 Describe the standardised metric and transformation methods used	Describe the standardised metric for each outcome. Explain why the metric(s) was chosen, and describe any methods used to transform the intervention effects, as reported in the study, to the standardised metric, citing any methodological guidance consulted	N/A	Not applicable; outcomes were reported descriptively as fracture union, time to union, and complications. No standardised effect metrics or transformations were applied (Methods – Data synthesis and reporting).
3 Describe the synthesis methods	Describe and justify the methods used to synthesise the effects for each outcome when it was not possible to undertake a meta-analysis of effect estimates	Data synthesis & eMethods 2	

4 Criteria used to prioritise results for summary and synthesis	Where applicable, provide the criteria used, with supporting justification, to select the particular studies, or a particular study, for the main synthesis or to draw conclusions from the synthesis (e.g., based on study design, risk of bias assessments, directness in relation to the review question)	Eligibility criteria, eTable 1, and eMethods 5	
5 Investigation of heterogeneity in reported effects	State the method(s) used to examine heterogeneity in reported effects when it was not possible to undertake a meta-analysis of effect estimates and its extensions to investigate heterogeneity	Data synthesis & eMethods 6	
6 Certainty of evidence	Describe the methods used to assess certainty of the synthesis findings	Data synthesis and Discussion	
7 Data presentation methods	Describe the graphical and tabular methods used to present the effects (e.g., tables, forest plots, harvest plots). Specify key study characteristics (e.g., study design, risk of bias) used to order the studies, in the text and any tables or graphs, clearly referencing the studies included	Study characteristics & Table 2	
Results			
8 Reporting results	For each comparison and outcome, provide a description of the synthesised findings, and the certainty of the findings. Describe the result in language that is consistent with the question the synthesis addresses, and indicate which studies contribute to the synthesis	SWiM analysis	
Discussion			
9 Limitations of the synthesis	Report the limitations of the synthesis methods used and/or the groupings used in the synthesis, and how these affect the conclusions that can be drawn in relation to the original review question	Discussion	

eMethods 3: Documentation of deviations to the pre-registered protocol

We have followed guidance provided by Moreau and Gamble (2020) on reporting and justifying deviations from the study protocol [2].

Moreau, D. & Gamble, B. (2022). Conducting a Meta-Analysis in the Age of Open Science. *Psychological Methods, 27* (3), 426-432.

Deviation (1) Expansion of electronic databases searched

Description	The original PROSPERO-registered protocol specified PubMed/MEDLINE as the sole electronic database for literature searching. During the conduct of the review, the search strategy was amended to include two additional databases: CINAHL and Scopus. All databases were searched from inception to December 2025 using the same core search terms and eligibility criteria.
Justification	Given the rarity of osteopoikilosis and the anticipated small number of eligible studies, the initial PubMed-only strategy was considered insufficient to ensure comprehensive case ascertainment. CINAHL was added to capture relevant case reports and clinical literature from allied health and nursing journals, while Scopus was included to enhance coverage of orthopedic, surgical, and international publications that may not be indexed in PubMed. Expanding the database coverage was therefore necessary to reduce the risk of incomplete retrieval and publication bias.
Impact	**Major**: The inclusion of CINAHL and Scopus increased the sensitivity of the search and resulted in the identification of additional eligible records that were not indexed in PubMed. Importantly, the amendment did not alter the predefined eligibility criteria, outcomes of interest, or data synthesis methods. As such, this deviation is unlikely to introduce bias and instead strengthens the completeness and validity of the systematic review.

eMethods 4 Search strategy

Search strategy on Pubmed/Medline

Description	Search terms
A comprehensive search was conducted in PubMed/MEDLINE from database inception to December 2025. The search strategy combined Medical Subject Headings (MeSH) and free-text keywords related to osteopoikilosis and fractures. No filters for study design were applied due to the anticipated rarity of eligible studies.	(“osteopoikilosis”[MeSH Terms] OR “osteopoikilosis”[Title/Abstract] OR “spotted bone disease”[Title/Abstract]) AND (“fracture”[MeSH Terms] OR “fractures”[Title/Abstract] OR “trauma”[Title/Abstract] OR “injury”[Title/Abstract] OR “bone healing”[Title/Abstract])

Search strategy on CINAHL

Description	Search terms
The CINAHL database was searched from inception to December 2025 to identify additional case reports and clinical literature not indexed in PubMed. Controlled vocabulary (CINAHL Headings) and free-text keywords were used.	((MH "Osteopoikilosis") OR "osteopoikilosis" OR "spotted bone disease") AND ((MH "Fractures+") OR fracture* OR trauma* OR injury* OR "bone healing")

Search strategy on Scopus

Description	Search terms
Scopus was searched from inception to December 2025 to enhance retrieval of orthopedic and international publications not consistently indexed in PubMed. Searches were conducted using title, abstract, and keyword fields.	(TITLE-ABS-KEY("osteopoikilosis" OR "spotted bone disease")) AND (TITLE-ABS-KEY("fracture" OR "fractures" OR "trauma" OR "injury" OR "bone healing"))

Search strategy on hand searching (Google/Google scholar)

Description	Search terms
Supplementary hand-searching was conducted to identify grey literature and non-indexed case reports.	“osteopoikilosis” fracture; “osteopoikilosis” trauma; “osteopoikilosis” bone healing; “spotted bone disease” fracture

eMethods 5 Modification of the Newcastle-Ottawa Scale

Rationale for modification

The original Newcastle–Ottawa Scale (NOS) was developed to assess the quality of non-randomized comparative studies, particularly cohort and case–control designs [3]. However, all studies included in the present systematic review consisted of single case reports or small case series without comparator groups. Consequently, several NOS domains (e.g., selection of non-exposed cohorts, comparability between groups, and control for confounding) were not applicable.

To ensure a transparent, reproducible, and appropriate assessment of methodological quality, we therefore applied a modified NOS framework tailored to descriptive clinical reports, in line with previous systematic reviews synthesizing rare diseases and fracture case literature.

Modified NOS domains and scoring

The modified NOS assessed study quality across four core domains relevant to case-based evidence: Selection, Ascertainment, Outcome assessment, and Follow-up adequacy. Each domain was rated using a binary or graded star-based system, with a maximum possible score of 5 stars.

Domain 1: Selection (maximum 2 stars)

This domain evaluated the clarity and validity of case identification.

- ★ Confirmed diagnosis of osteopoikilosis
  One star was awarded if the diagnosis was clearly established based on characteristic radiographic findings (bilateral, symmetric, periarticular sclerotic lesions) with or without supportive clinical or genetic information.
- ★ Clear fracture characterization
  One star was awarded if the fracture site, mechanism of injury, and diagnostic modality were clearly described.

Domain 2: Ascertainment of exposure and outcome (maximum 1 star)

- ★ Reliable assessment of fracture and management
  One star was awarded if fracture diagnosis and treatment were confirmed using objective measures (e.g., radiographs, CT, MRI) and the management strategy (operative or non-operative) was explicitly described.

Domain 3: Outcome assessment (maximum 1 star)

- ★ Adequate reporting of fracture healing outcome
  One star was awarded if fracture healing was assessed using radiographic evidence, clinical union, or both, with a clearly stated outcome (union, delayed union, or nonunion).

Domain 4: Follow-up adequacy (maximum 1 star)

- ★ Sufficient follow-up duration
  One star was awarded if the follow-up period was sufficient to reasonably assess fracture union based on fracture type and anatomical location (generally ≥6 weeks for upper extremity fractures and ≥12 weeks for lower extremity or weight-bearing fractures), or if complete union was explicitly documented.

Overall quality classification

Based on the total number of stars awarded, studies were classified as follows:

- High quality: 4–5 stars
- Moderate quality: 2–3 stars
- Low quality: 0–1 star

Quality assessment was performed independently by two reviewers. Any discrepancies were resolved through consensus discussion.

Role of quality assessment in data synthesis

Given the descriptive nature of the included studies and the absence of quantitative synthesis, study quality ratings were not used to weight outcomes statistically. Instead, they were incorporated into the SWiM qualitative synthesis to contextualize the robustness and consistency of reported fracture healing outcomes across studies.

eMethods 6. Justification of groupings of studies for SWiM

In accordance with the Synthesis Without Meta-analysis (SWiM) reporting guideline, studies were grouped a priori to facilitate a structured qualitative synthesis and to reduce conceptual heterogeneity across included reports.

Grouping by fracture location was performed based on anatomical and biomechanical considerations. Fractures were categorized into axial skeleton, upper extremity, and lower extremity fractures. This classification reflects clinically meaningful differences in load-bearing demands, fixation strategies, and biological healing environments. In particular, fractures involving the lower extremity represent a critical test of mechanical competence in osteopoikilotic bone, whereas upper extremity fractures are typically non–weight-bearing and biologically distinct. Grouping by location therefore allows contextual interpretation of fracture healing outcomes in relation to mechanical stress.

Grouping by treatment strategy was performed to distinguish between operative and non-operative management. This distinction is clinically relevant, as operative fixation directly challenges implant stability and bone–implant interface integrity in sclerotic bone, while non-operative management primarily reflects the intrinsic biological capacity for fracture repair. Separating these groups enabled evaluation of whether osteopoikilosis differentially affects healing outcomes depending on mechanical intervention.

These groupings were predefined, clinically driven, and applied consistently across studies. Given the absence of quantitative effect estimates and the descriptive nature of the included evidence, outcomes were synthesized narratively within each group without statistical pooling, in line with SWiM recommendations.

eTable 1 Eligibility criteria

Domain	Inclusion criteria	Exclusion criteria
Study design	Case reports, case series, observational studies (prospective or retrospective) reporting fractures in patients with osteopoikilosis	Reviews, editorials, commentaries without original patient data; animal or in vitro studies
Population	Human patients of any age or sex with a diagnosis of osteopoikilosis	Studies without confirmed osteopoikilosis diagnosis
Diagnosis of osteopoikilosis	Radiographic diagnosis based on characteristic symmetric sclerotic bone islands, with or without additional imaging (CT, MRI, bone scan, PET/CT)	Studies in which osteopoikilosis could not be distinguished from osteoblastic metastases or other sclerosing bone disorders
Condition of interest	Any traumatic fracture occurring in patients with osteopoikilosis	Non-fracture-related reports; isolated osteopoikilosis without fracture
Fracture characteristics	Fractures at any anatomical site, including axial and appendicular skeleton	Studies without clear description of fracture location or mechanism
Intervention / management	Operative or non-operative fracture management using standard orthopedic principles	Reports lacking information on fracture treatment
Outcomes	Reported fracture healing outcomes, including union, time to union, or qualitative assessment of healing	Absence of fracture healing or follow-up data
Publication type	Full-text articles, letter to the editor	Conference abstracts without sufficient clinical details
Language	English-language publications	Non-English publications
Duplicated data	The most complete report retained when multiple publications described the same patient	Duplicate reports of the same patient population without additional data

eTable 2 Applicability assessment using a ROBINS-E–informed framework

Appendix 1: framework for extending ROBINS-E to address appropriateness of studies to a review question[4]

Addressing appropriateness, Part I. (In advance) ***Specify the review question***		**Enter a response**
I.1 Population	Describe the types of individuals to whom the review question applies (e.g. general population, specific groups, employment status, age).	The population of interest comprises individuals of any age and sex with a confirmed diagnosis of osteopoikilosis, who sustained one or more skeletal fractures. Both pediatric and adult patients were considered eligible, reflecting the broad age range in which osteopoikilosis has been reported and fractures have been documented.
I.2 Exposure of interest	This is the factor whose causal effect on the outcomes of interest is to be addressed in the review.	The exposure of interest is osteopoikilosis, a rare sclerosing bone dysplasia characterized by multiple focal sclerotic bone islands, considered as a pre-existing skeletal condition potentially influencing fracture characteristics, management, and healing outcomes.
I.3 Exposure measures of interest	These are the ways in which the exposure of interest may be measured or assessed. They include both direct measures of exposure (e.g. personal radiation exposure measures), indirect measures of exposure (e.g. biomarkers) and proxies or surrogates (e.g. time spent in a uranium mine).	Exposure to osteopoikilosis was ascertained primarily through characteristic radiographic findings, including bilateral, symmetric, periarticular sclerotic lesions. Additional supportive measures included advanced imaging modalities (e.g. CT, DXA, 18-NaF PET/CT) or clinical documentation, where available. Genetic confirmation was not required.
I.4 Comparisons of interest	Comparisons may be of an exposed with an unexposed group, or across a range of exposure levels or intensities. At the review level, the answer to this question may sometimes be “any”.	No explicit comparison group was required. The review aimed to describe fracture characteristics and healing outcomes within osteopoikilotic patients, without direct comparison to unexposed populations. At the review level, comparisons were therefore considered “any” or not applicable.
I.5 Periods and patterns of exposure	Specify the exposure windows and patterns of exposure that are of interest in the review. For example, will the review be restricted to exposure during a fixed age range (or during pregnancy)? Is a cumulative, average or peak exposure of interest? At the review level, the answer to these questions may sometimes be “any”.	No restrictions were applied regarding the timing or duration of exposure. Osteopoikilosis is a congenital or developmental condition; therefore, exposure was assumed to be lifelong. All fractures occurring at any time point were considered eligible.

I.6	Outcomes of interest	Specify the outcomes that are of interest. An outcome is a health state, endpoint or marker on which the causal effect of the exposure is to be addressed in the review.	The primary outcomes of interest were fracture healing outcomes, including radiographic or clinical union, time to union, and reported complications such as delayed union, nonunion, implant failure, or fixation instability. Secondary outcomes included fracture location, management strategy, and follow-up duration.

Addressing appropriateness, Part II: (In advance) ***Address appropriateness of studies to the review question***		**Enter a response**
II.1	Will the review, and therefore risk of bias assessments, be restricted to particular study designs? If so specify these.	The review was restricted to case reports and small case series reporting original patient data. Observational comparative studies were not available. Reviews, editorials, conference abstracts, and studies without individual-level fracture data were excluded.
II.2	Will the review, and therefore risk of bias assessments, be restricted to studies of individuals who were exposed during a specified period ("exposure window") considered to be necessary to detect a relevant effect of the exposure? If so, specify this period (or periods), and whether it (they) relate(s) to specific outcomes.	No restriction was applied regarding exposure windows. As osteopoikilosis is a non-time-varying skeletal condition, all studies reporting fractures in patients with osteopoikilosis were considered applicable regardless of age at fracture.
II.3	Will the review, and therefore risk of bias assessments, be restricted to studies in which there is a minimum range of exposure levels considered to be necessary to detect a relevant effect of the exposure? If so, specify this range (or ranges), and whether it (they) relate(s) to specific outcomes.	No minimum range or gradation of exposure severity was required. Osteopoikilosis was treated as a binary exposure (present/absent), as severity grading was inconsistently reported across studies.
II.4	Will the review, and therefore risk of bias assessments, be restricted to studies in which there is a minimum follow-up period (from start of exposure) considered to be necessary to detect a relevant effect of the exposure? If so, specify this period (or periods), and whether it (they) relate (s) to specific outcomes.	A minimum follow-up duration was not predefined; however, studies were required to report fracture healing status or outcome sufficient to reasonably assess union for the fracture type involved. Studies lacking outcome information were excluded.
II.5	Will the review, and therefore risk of bias assessments, be restricted to particular types of exposure-outcome relationship ('dose-response' models)? If so specify these (appropriate models may vary between outcomes).	No specific dose–response or exposure–outcome models were prespecified. Given the descriptive nature of the evidence and absence of quantitative exposure metrics, outcomes were synthesized narratively without assumptions of linear or non-linear relationships.

eTable 3: Table of studies excluded using ROBINS-E tool

Study (Author, Year)	Reason for exclusion
Jonasch[5]**, 1956**	Unable to reliably extract outcome data due to language barrier (German)
Lagier[6]**, 1984**	No fracture healing outcome reported
Sim E[7]**, 1989**	Unable to reliably extract outcome data due to language barrier (German)
Mochul'skii[8]**, 1992**	Unable to reliably extract outcome data due to language barrier (Russian)
Zahar[9]**, 2002**	Unable to reliably extract outcome data due to language barrier (French)
Rosa-Juana[10]**, 2024**	No fracture healing outcome reported
Fidan[11]**, 2025**	No clinical follow-up

eTable4: The Modified Newcastle Ottowa Scale quality assessment

Author/date	Selection	Ascertainment	Outcome	Follow-up	Study Quality[a]
Gurcan[12], 2012	★★	★	★	★	High
Du Mortier[13], 2014	★★	★	★	★	High
Khattak[14], 2012 **Ali[15], 2017[b]**	★	★	★	-	Moderate
Liu[16], 2024	★	★	★	-	Moderate
Pollen[17], 1968	★	★	★	★	High
Spiezia[18], 2025	★★	★	★	★	High
Buyukbebeci[19], 2005	★★	★	★	★	High
Ye[20], 2017	★★	★	★	★	High
Bansal[21], 2013	★★	★	★	★	High
Ogalde-Bravo[22], 2026	★★	★	★	★	High
Current Case Suwanaratana, 2025	★★	★	★	★	High

[a] Study quality categories were used to contextualize findings within the qualitative synthesis and were not used for statistical weighting.

[b] Khattak et al. (2012) and Ali (2017) describe the same clinical case and were synthesized as a single patient.

eReferences

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