

INPLASY

Clinical and surgical outcomes in pediatric meningiomas: protocol of a systematic review and meta-analysis

INPLASY2026100104

doi: 10.37766/inplasy2026.10.0104

Received: 25 October 2026

Published: 26 October 2026

Bolt, AJ; Gupta, K; Gupta, K; Vadset, T; Kaur, B; John, J; Liriano, G; Agarwal, V; Kobets, AJ.

Corresponding author:

Khushal Gupta

khushal.gupta@einsteinmed.edu

Author Affiliation:

Leo M. Davidoff Department of Neurological Surgery, Montefiore Medical Center, Albert Einstein College of Medicine, Bronx, New York, USA.

ADMINISTRATIVE INFORMATION

Support - None. This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors.

Review Stage at time of this submission - Completed but not published.

Conflicts of interest - None declared.

INPLASY registration number: INPLASY2026100104

Amendments - This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 26 October 2026 and was last updated on 26 October 2026.

INTRODUCTION

Review question / Objective Review question: in children and adolescents with intracranial or spinal meningiomas, what are the clinical and surgical characteristics and outcomes reported in the contemporary literature, and which factors are associated with recurrence and death?

Primary objective: to estimate pooled proportions of male sex, neurofibromatosis type 2 (NF2), WHO grade II–III histology, gross-total resection, adjuvant radiotherapy or radiosurgery, recurrence or progression, and mortality.

Secondary objective: to assess, in within-study (two-arm) comparisons, extent of resection, WHO grade, NF2 status and adjuvant radiotherapy as factors associated with recurrence or progression, and extent of resection and WHO grade as factors associated with death.

PICOS: population, patients aged 18 years or younger with meningiomas; exposures, the factors listed above; comparators, the complementary group within the same study; outcomes, the seven proportions and risk ratios for recurrence or progression and for death; study designs, retrospective cohort studies, case series and institutional registries with at least five pediatric patients.

Rationale Meningiomas are rare in children and adolescents, accounting for fewer than 5% of pediatric central nervous system tumors, and their clinicopathological and prognostic features are less well characterized than those of adult meningiomas. The evidence consists almost entirely of small, retrospective single-institution series that differ in their populations, calendar periods, definitions of extent of resection and recurrence, and length of follow-up, so the reported rates of high-grade histology, gross-total resection, adjuvant treatment, recurrence and mortality vary widely. Synthesis is further

complicated by repeated reports from the same institutions and by cohorts assembled by the treatment received. No dedicated guideline for pediatric meningiomas is available from the AANS and CNS. A systematic review and meta-analysis that resolves overlapping cohorts and handles treatment-selected series separately can provide quantitative benchmarks, and an estimate of the factors associated with recurrence and death, to inform counseling, surgical planning and surveillance.

Condition being studied Meningiomas in patients aged 18 years or younger. Pediatric meningiomas are uncommon tumors arising from the meninges of the brain or spinal cord. Compared with adult meningiomas, they are reported to show a less pronounced female predominance, a higher proportion of WHO grade II–III (atypical and anaplastic) histology, a stronger association with neurofibromatosis type 2 and a different anatomical distribution. Treatment is primarily surgical, with radiotherapy or radiosurgery reserved for residual, recurrent, surgically inaccessible or higher-grade tumors. Outcomes of interest include extent of resection, recurrence or progression, and death.

METHODS

Search strategy The same two-concept structure (meningioma AND pediatric population) was used in every database, combining controlled vocabulary where available with free-text terms. No outcome, study-design or language terms were included; these were applied at screening.

PubMed (MEDLINE), searched 7 September 2026, limited to September 2016 to September 2026: (Meningioma[Mesh] OR meningioma*[tiab]) AND (Child[Mesh] OR Adolescent[Mesh] OR Infant[Mesh] OR Pediatrics[Mesh] OR child*[tiab] OR pediatric*[tiab] OR paediatric*[tiab] OR adolescen*[tiab] OR juvenil*[tiab] OR infan*[tiab] OR teen*[tiab] OR youth*[tiab] OR boys[tiab] OR girls[tiab] OR schoolchild*[tiab])

Embase ([Embase.com](https://www.embase.com)), searched 7 September 2026, limited to publication years 2016–2026: ('meningioma'/exp OR meningioma:ti,ab,kw OR meningiomas:ti,ab,kw OR 'meningeal neoplasm':ti,ab,kw OR 'meningeal neoplasms':ti,ab,kw OR 'meningeal tumor':ti,ab,kw OR 'meningeal tumour':ti,ab,kw OR 'meningeal tumors':ti,ab,kw OR 'meningeal tumours':ti,ab,kw) AND ('child'/exp OR 'adolescent'/exp OR 'infant'/exp OR 'pediatrics'/exp OR 'juvenile'/exp OR child*:ti,ab,kw OR pediatric*:ti,ab,kw OR

paediatric*:ti,ab,kw OR adolescen*:ti,ab,kw OR juvenile*:ti,ab,kw OR infan*:ti,ab,kw OR teen*:ti,ab,kw OR youth*:ti,ab,kw OR boys:ti,ab,kw OR girls:ti,ab,kw OR schoolchild*:ti,ab,kw)

Web of Science Core Collection, searched 7 September 2026, limited to September 2016 to September 2026:

TS=(meningioma*) AND TS=(child* OR pediatric* OR paediatric* OR adolescen* OR juvenil* OR infan* OR teen* OR youth* OR boys OR girls OR schoolchild*)

Cochrane Library (CENTRAL), searched 30 September 2026, limited to publication years 2015–2026:

([mh Meningioma] OR (meningioma*):ti,ab,kw) AND ([mh Child] OR [mh Adolescent] OR [mh Infant] OR [mh Pediatrics] OR (child* OR pediatric* OR paediatric* OR adolescen* OR juvenile* OR infan* OR teen* OR youth* OR boys OR girls OR schoolchild*):ti,ab,kw)

Total records retrieved: 5,483.

Participant or population Patients aged 18 years or younger at diagnosis or surgery with histologically or radiologically confirmed intracranial or spinal meningiomas. Studies using an upper age cut-off of 20 or 21 years were eligible and were flagged. Outcomes had to be reported for the pediatric population separately from any adult population. Cohorts assembled by the treatment received (radiosurgery or proton therapy series) were eligible and were flagged as treatment-selected.

Intervention Not applicable to the pooled proportions, which describe the cohort, tumor, treatment and outcome characteristics of pediatric meningiomas. For the comparative analyses, the exposures (prognostic factors) were subtotal or partial resection, WHO grade II–III histology, NF2-associated disease and adjuvant radiotherapy.

Comparator For the comparative analyses, the complementary group within the same study: gross-total resection, WHO grade I histology, sporadic (non-NF2) disease and no adjuvant radiotherapy. Not applicable to the pooled proportions.

Study designs to be included Retrospective cohort studies, case series and institutional registries reporting at least five pediatric patients with meningiomas. Case reports, series of fewer than five pediatric patients, conference abstracts, editorials, letters, narrative reviews, and preclinical

or purely molecular studies without clinical outcomes were excluded.

Eligibility criteria Inclusion: patients aged 18 years or younger at diagnosis or surgery (studies using a cut-off of 20 or 21 years were included and flagged); histologically or radiologically confirmed intracranial or spinal meningiomas; at least one pre-specified outcome reported for the pediatric population separately from any adult population; at least five pediatric patients; full text in English.

Excluded from pooling on methodological grounds: cohorts selected by the presence of NF2; national administrative databases, whose patients cannot be assumed independent of the institutional series; mixed-age or mixed-tumor series without separate pediatric data; and cohorts selected by the occurrence of recurrence. Cohorts assembled by the treatment received were eligible but flagged as treatment-selected.

Overlapping cohorts: institutional cohorts reported more than once were identified by center, period and patient count, and a single index study, in general the report with the largest cohort for the outcome in question, was retained for each.

Information sources PubMed (MEDLINE), Embase ([Embase.com](https://www.embase.com)) and the Web of Science Core Collection, searched on 7 September 2026, and the Cochrane Central Register of Controlled Trials (CENTRAL, Cochrane Library), searched on 30 September 2026. PubMed and the Web of Science were limited to September 2016 to September 2026, Embase to publication years 2016–2026 and CENTRAL to 2015–2026. Trial registers and preprint servers were not searched directly.

Main outcome(s) Seven pooled proportions, each calculated with the denominator reported for that item:

1. Male sex, among patients with sex reported.
2. Neurofibromatosis type 2 (NF2), among patients with the item reported.
3. High-grade histology: WHO grade II and III tumors among graded tumors, as graded by each study under the WHO edition in use.
4. Gross-total resection: Simpson grade I–II resection, or complete or R0 resection by the study's own definition, among patients operated on.
5. Adjuvant radiotherapy or radiosurgery (postoperative radiotherapy, stereotactic radiosurgery or proton therapy), among patients with the item reported.

6. Recurrence or progression: radiological or clinical recurrence or progression during follow-up, with the denominator used by each study.

7. Mortality: deaths from any cause (unless a study specified disease-related deaths), among patients.

Additional outcome(s) Risk ratios from within-study (two-arm) comparisons: subtotal or partial versus gross-total resection, WHO grade II–III versus grade I, NF2-associated versus sporadic disease, and adjuvant radiotherapy versus none, for recurrence or progression; and extent of resection and WHO grade for death.

Added after the primary analyses: tumor location (classified as skull base or other), preoperative embolization, causes of death and thrombotic or thromboembolic complications, summarized descriptively; and a seventh comparison, skull base versus other intracranial location as a factor associated with incomplete (subtotal or partial) resection.

Data management Records were imported into Rayyan and deduplicated. For PubMed, Embase and the Web of Science, a documented rule-based pass removed conference abstracts, records not concerning meningiomas, records with no pediatric-population signal, single case reports, editorials and letters, non-English publications and preclinical work, with the reason recorded for every record; the pass was validated by confirming that every included study survived it. The remaining records, including the unique CENTRAL records, were screened on title and abstract by two reviewers working independently, with disagreements resolved by consensus. Rayyan's relevance ranking was used to order records for screening and to re-check the excluded pool; no record was excluded on the basis of the ranking alone. Full texts were divided among three reviewers for eligibility assessment, and data were extracted by five reviewers into a structured workbook against a pre-specified data dictionary. Items not reported were coded NR, and the study was omitted from that outcome rather than assigned zero. Tumor-level counts were flagged and converted to patient-level counts where possible. Record-level screening decisions and the extraction workbook are held by the corresponding author.

Quality assessment / Risk of bias analysis

Methodological quality for the pooled proportions was appraised with the Joanna Briggs Institute Critical Appraisal Checklist for Case Series (ten items, each answered Yes, No or Unclear, with the supporting text recorded). The comparison of

subtotal or partial versus gross-total resection for recurrence or progression was appraised with ROBINS-I in every contributing study, each of the seven domains judged Low, Moderate, Serious or Critical, with the overall judgment taken from the worst domain. Judgments were made from the full text, with uncertainty recorded as Unclear. Certainty of evidence was not formally graded with GRADE; because all estimates derive from retrospective, non-randomized cohorts, they are regarded as of low or very low certainty.

Strategy of data synthesis Each prevalence outcome was pooled as a single-arm proportion with a random-effects model (inverse-variance method, DerSimonian–Laird estimator) after Freeman–Tukey double-arcsine transformation, and back-transformed for presentation, with 95% confidence intervals, I^2 , τ^2 , Cochran's Q and 95% prediction intervals.

For the comparative analyses, 2×2 tables were built within each study (never by combining arms across studies), with the index arm defined a priori as the arm hypothesized to do worse, and risk ratios were pooled with a random-effects inverse-variance model (DerSimonian–Laird). A comparison was pooled only if at least five studies contributed and at least one event occurred; in studies with a zero cell, 0.5 was added to the events and to the number of patients in each arm, and estimates resting entirely on the continuity correction were reported as direction only. Comparisons with fewer than five studies were reported narratively.

Small-study effects were examined with funnel plots and Egger's test. Tumor location, embolization, causes of death and thrombotic complications were summarized descriptively. Analyses were run in MetaAnalysisOnline.

Subgroup analysis No subgroup analysis or meta-regression was performed; between-study heterogeneity was explored through the sensitivity analyses.

Sensitivity analysis Six sensitivity analyses were performed. The exclusion of treatment-selected cohorts was planned before pooling; the other five were added during the analysis. For the pooled proportions: excluding Shanahan 2026, which has a possible but unquantifiable cohort overlap with Samanci 2023; excluding the two radiotherapy-selected cohorts from the adjuvant radiotherapy outcome; excluding all four treatment-selected cohorts (surgery-only estimate); excluding studies with fewer than ten patients; and excluding the one included study that could not be appraised for risk

of bias. For the comparative analysis, the resection–recurrence comparison was repeated after removing studies at critical risk of bias on ROBINS-I.

Language restriction English only.

Country(ies) involved United States.

Other relevant information Retrospective registration. This review was not registered before it began; this record is submitted after the review was completed and before journal submission. Searches were run on 7 September 2026 (PubMed, Embase, Web of Science) and 30 September 2026 (CENTRAL), and screening, data extraction, risk-of-bias assessment and analyses were completed before this registration. The eligibility criteria and outcome definitions were fixed in a written data dictionary before data extraction.

Decisions made after results were available: five of the six sensitivity analyses were added during the analysis, and, after the primary analyses, four further data items (tumor location, preoperative embolization, causes of death and thrombotic complications) were extracted and the comparison of skull base versus other location for incomplete resection was added. These are identified as post hoc in the review, which is reported according to PRISMA 2020.

Keywords Pediatric meningiomas; meta-analysis; extent of resection; skull base; WHO grade; neurofibromatosis type 2; recurrence; mortality.

Dissemination plans Publication in a peer-reviewed journal. Study-level data are reported in the article and its supplementary material, and the extraction workbook and record-level screening decisions are available from the corresponding author on reasonable request.

Contributions of each author

Author 1 - Anne J. Bolt - Conception and design; literature screening, data extraction and risk-of-bias appraisal; drafting of the manuscript.

Author 2 - Khushal Gupta - Conception and design; literature screening, data extraction and risk-of-bias appraisal; statistical analysis; drafting of the manuscript.

Author 3 - Kasmira Gupta - Conception and design; literature screening, data extraction and risk-of-bias appraisal; drafting of the manuscript.

Author 4 - Taylor Vadset - Conception and design; literature screening, data extraction and risk-of-bias appraisal; critical revision of the manuscript.

Author 5 - Barnit Kaur - Conception and design; literature screening, data extraction and risk-of-bias appraisal; critical revision of the manuscript.

Author 6 - Jenny John - Conception and design; literature screening, data extraction and risk-of-bias appraisal; critical revision of the manuscript.

Author 7 - Genesis Liriano - Conception and design; supervision; critical revision of the manuscript.

Author 8 - Vijay Agarwal - Conception and design; supervision; critical revision of the manuscript.

Author 9 - Andrew J. Kobets - Conception and design; supervision; critical revision of the manuscript.