INPLASY

Hemophilic pseudotumor of the maxilla secondary to endodontic treatment: case report and systematic review

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ADMINISTRATIVE INFORMATION

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

Conflicts of interest - None declared.

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Amendments - This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 13 October 2025 and was last updated on 13 October 2025.

INTRODUCTION

Review question / Objective In patients with hemophilia (P), what clinical, radiographic, and pathological characteristics (O) have been reported in cases of hemophilic pseudotumor involving the maxilla (I), compared with other craniofacial locations or triggering factors such as dental procedures or trauma (C), according to case reports and case series (S)?

Rationale Hemophilic pseudotumor (HP) is a rare but severe complication of hemophilia, characterized by progressive bleeding and bone destruction. Although frequently described in long bones and the mandible, involvement of the maxilla is exceptionally uncommon, with only a limited number of cases reported. Given this rarity and the potential for misdiagnosis as odontogenic or neoplastic lesions, a systematic review was conducted to identify and synthesize all published cases of HP affecting the maxilla.

Condition being studied Hemophilic pseudotumor (HP) — a chronic, encapsulated hemorrhagic lesion resulting from recurrent bleeding in patients with hemophilia or related coagulopathies, localized in this review to the maxillary bone.

METHODS

Search strategy Searches were performed in PubMed, Scopus, and ScienceDirect for indexed literature and Google Scholar for gray literature (up to September 2024).
Example (PubMed):
("Pseudotumor, Hemophilia" [MeSH]) AND ("Hemophilic pseudotumor") AND ("Hemophilic cysts") AND ("Haemophilic pseudotumor") AND

("Maxilla" OR "Gnathic") NOT ("Mandibule") NOT ("Animal").
Equivalent Boolean strings were adapted for each database.

Participant or population Patients of any age or sex diagnosed with hemophilia A, B, or C, or other hemorrhagic diatheses (e.g., von Willebrand disease), who developed pseudotumor lesions in the maxilla confirmed by clinical, radiographic, and histopathological evaluation.

Intervention No therapeutic intervention was applied by the reviewers; the exposure of interest was the presence of hemophilia and potential triggering events such as dental procedures, trauma, or spontaneous bleeding episodes leading to pseudotumor formation.

Comparator Not applicable in the experimental sense. When reported, comparisons were drawn between maxillary vs. mandibular lesions or between cases with and without identifiable triggering events (e.g., dental treatment, trauma).

Study designs to be included Case reports and case series describing HP involving the maxilla were included. Systematic reviews without original cases, editorials, conference abstracts, and letters were excluded.

Eligibility criteria
Inclusion:
- Reports providing sufficient clinical and histological data to confirm HP diagnosis and anatomical location in the maxilla.
- Publications in any language and from any year.
Exclusion:
- Reports without confirmatory histopathology or incomplete localization data.
- Animal studies, book chapters, and editorials.

Information sources Electronic databases: PubMed, Scopus, ScienceDirect.
Gray literature: Google Scholar.
All sources were searched up to September 2024.

Main outcome(s) To determine the clinical, radiographic, and histopathological characteristics of hemophilic pseudotumor in the maxilla, including patient demographics, type and severity of hemophilia, precipitating factors, and treatment outcomes.

Additional outcome(s)
Secondary outcomes included:
- Geographic distribution of reported cases.
- Treatment modalities (surgery, replacement therapy, radiotherapy).
- Follow-up duration and recurrence rates.

Data management All retrieved articles were screened for duplicates. Titles and abstracts were assessed for eligibility, and full texts were reviewed when relevant. Data extraction (authors, year, demographics, site, treatment, follow-up, hemophilia type/severity) was entered into a descriptive database for synthesis.

Quality assessment / Risk of bias analysis The Joanna Briggs Institute (JBI) critical appraisal tools for case reports and case series were applied.
Each study was scored as Yes/No/Unclear, and bias levels were categorized as high (< 50%), moderate (50–69%), or low (≥ 70%).

Strategy of data synthesis A descriptive qualitative synthesis was performed. Quantitative pooling (meta-analysis) was not feasible due to the heterogeneity and rarity of reported cases. Data were summarized in tabular form and analyzed using descriptive statistics (counts, ranges, and percentages).

Subgroup analysis Comparisons were qualitatively explored by age group, sex, hemophilia type/severity, and treatment modality, emphasizing clinical patterns and outcomes.

Sensitivity analysis Not applicable; no quantitative synthesis was conducted. Sensitivity was implicitly assessed by the JBI quality classification and comparison of findings between high- and low-bias reports.

Language restriction No language restrictions were applied; reports in all languages were eligible provided sufficient diagnostic detail was available.

Country(ies) involved The review was conducted by authors affiliated with institutions in Mexico (Universidad de Guadalajara, IMSS, UAEMex).

Other relevant information The study adhered to PRISMA and CARE guidelines.
Institutional approval was obtained (University of Guadalajara, CI-07725).

Keywords hemophilic pseudotumor; hemophilia; maxilla; oral pathology; endodontic complication; case report; systematic review.

Dissemination plans The findings are intended for publication in a peer-reviewed dental or oral pathology journal (current submission: Dentistry

Journal, MDPI). Further dissemination will include presentations in hematology and oral pathology scientific meetings.

Contributions of each author

Author 1 - Jose Rodolfo Quiroz-Gomez - Contributed to data curation, formal analysis, and investigation of included studies. Participated in methodology design, project administration, and visualization of figures and tables. Drafted the original manuscript and participated in review and editing of the final version.
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Author 2 - Carlos Manuel Roa-Encarnación - Participated in the conceptualization of the study, contributed to investigation and resource acquisition, and assisted in validation of clinical and diagnostic data.
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Author 6 - Mario Nava-Villalba - Led the conceptualization and overall methodological design of the study. Responsible for formal analysis, validation, and supervision. Managed project administration, coordinated resources, and contributed to funding acquisition. Co-wrote the original draft and oversaw final review and editing of the manuscript.
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