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Branchial Cleft Anomalies as an Overlooked Etiology of Life-threatening Cervicothoracic Necrotizing Infections: A Systematic Review and Clinical Management Algorithm(Protocol)

Yuan, HW; Huang, SL; Teng, YS; Chen, YC; Yang, H; Xu, Y; Pan, HG.

Corresponding author:
Huwei Yuan

108849593@qq.com

Author Affiliation:
Department of Otorhinolaryngology, Shenzhen Children's Hospital.

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 10 April 2026 and was last updated on 10 April 2026.

INTRODUCTION

Review question / Objective This systematic review aims to evaluate the clinical characteristics, infection dissemination patterns, and management strategies of life-threatening cervicothoracic necrotizing infections (such as necrotizing fasciitis, descending necrotizing mediastinitis, and empyema) caused by branchial cleft anomalies (BCAs). The objective is to highlight BCAs as an overlooked etiology in these severe infections and to propose a standardized, five-phase clinical management algorithm.

Rationale Deep neck infections can rapidly progress to life-threatening conditions like necrotizing fasciitis and descending necrotizing mediastinitis. While odontogenic and tonsillar sources are well-recognized, branchial cleft anomalies (BCAs) are often overlooked as a primary etiology, especially in adults. Delayed recognition of a BCA-related infection can lead to inappropriate initial management, recurrent infections, and catastrophic outcomes. A systematic synthesis of existing literature is needed to elucidate the unique clinical trajectory of these cases and establish a definitive management algorithm.

Condition being studied Life-threatening cervicothoracic necrotizing infections, specifically necrotizing fasciitis, descending necrotizing mediastinitis, mediastinal abscess, and empyema, originating from congenital branchial cleft anomalies (including pyriform sinus fistulas, third, and fourth branchial anomalies).

METHODS

Search strategy PubMed, Embase, and Web of Science (Core Collection) were systematically searched from inception to December 2025. The Cochrane Library was also searched. The search strategy combined Concept 1 (branchial cleft anomalies) and Concept 2 (severe necrotizing

complications).For PubMed:("branchial cleft"[Title/Abstract] OR "branchial anomaly"[Title/Abstract] OR "branchial anomalies"[Title/Abstract] OR "branchial arch"[Title/Abstract] OR "branchial pouch"[Title/Abstract] OR "pyriform sinus fistula"[Title/Abstract] OR "pyriform fossa fistula"[Title/Abstract] OR "piriform sinus fistula"[Title/Abstract] OR "third branchial"[Title/Abstract] OR "fourth branchial"[Title/Abstract])AND("necrotizing fasciitis"[Title/Abstract] OR "necrotising fasciitis"[Title/Abstract] OR "necrotizing mediastinitis"[Title/Abstract] OR "descending mediastinitis"[Title/Abstract] OR "mediastinal abscess"[Title/Abstract] OR "empyema"[Title/Abstract] OR "pyothorax"[Title/Abstract] OR "life-threatening"[Title/Abstract] OR "fatal"[Title/Abstract] OR "death"[Title/Abstract] OR "necrotizing soft tissue infection"[Title/Abstract] OR "NSTI"[Title/Abstract]).

Participant or population Patients of any age presenting with a life-threatening cervicothoracic necrotizing infection (necrotizing fasciitis, descending necrotizing mediastinitis, mediastinal abscess, or empyema) in whom a branchial cleft anomaly was definitively identified as the primary source of infection.

Intervention Emergency surgical interventions (e.g., radical debridement, cervical drainage, video-assisted thoracoscopic surgery) and definitive treatment of the underlying branchial anomaly (e.g., open surgical excision, endoscopic cauterization).

Comparator Not applicable. This is a systematic review of case reports and case series without a specific comparator group.

Study designs to be included Case reports, case series, and observational studies.

Eligibility criteria Inclusion criteria: (1) definitive identification of a branchial cleft anomaly as the primary source of infection; (2) infection complicated by at least one life-threatening condition (necrotizing fasciitis, descending necrotizing mediastinitis, mediastinal abscess, empyema, airway compromise, or death); (3) sufficient clinical data available.Exclusion criteria: Uncomplicated BCA infections, reviews/guidelines without original case data, and studies where BCA was not confirmed as the primary source.

Information sources Electronic databases (PubMed, Embase, Web of Science Core Collection, Cochrane Library) and manual screening of reference lists of included studies and relevant review articles.

Main outcome(s) Patient demographics, type and laterality of the branchial cleft anomaly, nature of the life-threatening complication (e.g., necrotizing fasciitis, mediastinitis), and the extent of infection dissemination (e.g., mediastinal involvement rate).

Additional outcome(s) Microbiological findings, emergency surgical interventions performed, definitive treatment strategies for the underlying anomaly, recurrence rates, and patient mortality.

Data management Two independent reviewers screened titles, abstracts, and full texts. Discrepancies were resolved by consensus or consultation with a third reviewer. Data were extracted into a standardized spreadsheet.

Quality assessment / Risk of bias analysis The methodological quality of the included studies was assessed using the Joanna Briggs Institute (JBI) Critical Appraisal Checklist for Case Reports.

Strategy of data synthesis Due to the nature of the included studies (predominantly case reports) and the heterogeneity of the data, a qualitative narrative synthesis was performed. Clinical characteristics, surgical interventions, and outcomes were summarized descriptively and presented in tabular format.

Subgroup analysis Not applicable due to the limited number of cases.

Sensitivity analysis Not applicable.

Language restriction No language restrictions were applied.

Country(ies) involved China.

Other relevant information This systematic review proposes a novel five-phase clinical management algorithm for BCA-related life-threatening cervicothoracic infections.

Keywords Branchial cleft anomaly; necrotizing fasciitis; descending necrotizing mediastinitis; pyriform sinus fistula; algorithm.

Dissemination plans The findings will be submitted for publication in a peer-reviewed scientific journal.

Contributions of each author

Author 1 - Huwei Yuan - Conceived the study; jointly conducted double-person literature search, screening, quality control and disagreement resolution with B; performed data extraction/ analysis; drafted the manuscript; supervised the project and handled correspondence.
Email: 108849593@qq.com
Author 2 - Shuling Huang - Jointly conducted double-person literature work and disagreement resolution with A; verified data; participated in data analysis and manuscript revision.
Email: huang201071@126.com
Author 3 - Yishu Teng - Assisted in search strategy, literature/data sorting and manuscript revision.
Email: tys118@163.com
Author 4 - Yongchao Chen - Responsible for statistical analysis; assisted in chart preparation and paper review.
Author 5 - Hong Yang - Provided clinical/ pathological expert opinions; participated in discussions and manuscript revision.
Author 6 - Yang Xu - Assisted in evidence synthesis, reference sorting and final manuscript review.
Author 7 - Hongguan Pan - Provided senior supervision; critically revised the manuscript; reviewed the final version and provided funding.