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MULTIDISCIPLINARY MANAGEMENT OF MAJOR CONGENITAL ANOMALIES IN CHILDREN: SURGICAL AND LONGITUDINAL MEDICAL PERSPECTIVES — A SYSTEMATIC REVIEW AND EXPLORATORY META-ANALYSIS

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ABSTRACT

Focused care around major congenital anomalies in young people is well-documented to be long-term following surgical correction. The trajectory of management frequently requires several different perspectives: pediatric surgeons, medical subspecialists, rehabilitation teams, allied health practitioners, and psychosocial interventions. Although there is universal consensus about the importance of providing multidisciplinary care of congenital anomalies, the published evidence is fragmented within categories including anomaly, outcome, and clinic structure. This systematic review and exploratory meta-analysis pooled evidence from available evidence concerning multimodal management models for pediatric congenital anomalies that demand a coordinated surgical and medical follow-up. A PRISMA 2020 based systematic review was performed. The search of the literature ran from inception to 2026-03-01 in PubMed (MEDLINE), Embase, Scopus, Web of Science Core Collection, PsycINFO, and Cochrane CENTRAL was conducted, with structured grey-literature searches included. No language limitations were applicable. Two reviewers screened studies independently, extracted data across articles and discussed risk of bias, and another reviewer resolved potential disagreements. Studies eligible assessed multidisciplinary clinics, integrated care pathways, or structured longitudinal follow-up programs for children with congenital anomalies that required both operative and medical management. Results: Primary outcomes included hospital use, active management change after multidisciplinary review, and clinically significant care-process outcomes. Synthesis of random-effects was pre-expected as there was a pronounced degree of heterogeneity which led to limited quantity pooling to exploratory analyses of data with direct correlations. qualitative synthesis comprised 17 studies. The evidence base consisted of retrospective, single-center observational studies from tertiary settings. Across anomaly cohorts, multidisciplinary care models consistently demonstrated recognizing ongoing needs in addition to the index operation, improved

coordinated therapeutic decision-making, and was effective at minimising fragmented follow-up. Comparative studies indicated directional reductions in admissions, hospital days, or reactive procedures after the introduction of structured multidisciplinary clinics, but the limited and heterogeneous coverage in pooled comparative data limited these robust estimates. There was moderate to serious risk for bias in the majority of studies and generally a low level of certainty. Available evidence argues for multidisciplinary, longitudinal congenital anomaly care as a clinically congruent approach to children's health who need combined treatment and surveillance, rehabilitation and family-based care. Nevertheless, stronger multicenter prospective studies employing standard defined outcome measures are needed to draw firm causal inferences.

KEYWORDS:

1. INTRODUCTION

Congenital anomalies are still one of the most prevalent causes of infant mortality, childhood disability, re-hospitalization, and long-term health care utilization worldwide [1,2]. Current advances in neonatal intensive care, perioperative guidance, prenatal diagnosis, pediatric anesthesia and reconstructive techniques have vastly increased survival in many congenital disorders; however, survival gains have brought the clinical burden into chronic morbidity, developmental outcomes, functional impairment and prolonged health-service dependency during early and late childhood and adolescence [3–6]. In several groups of anomalies, cleft lip and palate, spina bifida, esophageal atresia/tracheoesophageal fistula (EA/TEF), anorectal malformations, congenital diaphragmatic hernia, and vascular anomalies, the first operation is merely a component of an extended path to care [4-9].

It is very infrequent to meet expectations of this long-term trajectory within a specialty. Speech outcomes following cleft repair rely on the collaboration of surgical, orthodontic, otolaryngologic, audiologic and speech-language assessments; bowel and bladder preservation in spina bifida is dependent on a comprehensive neurosurgical, urological, orthopedic, rehabilitation and psychosocial approach; aerodigestive morbidity after EA surgery often requires multi-disciplinary surgical, gastroenterological, respiratory, nutritional and airway assessments; and vascular anomalies often require a multidisciplinary, combined radiologic, dermatologic, pharmacologic and operative approach to decision making [5-12]. As a result multidisciplinary delivery has become the accepted organisational model in specialized congenital services.

Nevertheless, acceptance of multi-disciplinary management in its entirety is not evidence of effectiveness. The concept of "multidisciplinary" is also variable in the literature between a one-stop clinic, a shared case-review board, formal longitudinal follow-up programs, and a combination of consultation models [8-13]. Consequences are heterogeneous in nature too: management recommendations and referral patterns, hospitalization, speech-related outcomes, airway morbidity, burden to the family and value of services. Consequently, the literature is clinically convincing, yet methodologically challenging to synthesize.

This dilemma has real-world consequences. Multidisciplinary congenital clinics need coordinating scheduling, organizational support, dedicated staff and, in a lot of cases, a major financial and logistical investment. Such services may be considered integral to clinicians, but health systems are urgently demanding evidence that integrated care leads to better outcomes, avoids preventable admissions, prevents fragmentation and promotes timely intervention [10-14]. Success, too, may be evaluated by families as clarity of planning, less duplication, coordinated appointments and fewer missed developmental opportunities, and certainly not by the technical outcomes of surgery.

The review, therefore, presents clinical management of significant congenital anomalies in children; a multidisciplinary approach of surgical and medical management in areas in which long-term medical care is integrated with operating procedures. The goal was to compile evidence on integrated care models, to determine the extent to which quantitative pooling is possible for selected outcomes and to discover the structural and methodological characteristics shaping the field presently [5-14].

2. METHODS

2.1. Study Design and Reporting Framework

The present study was conducted as a systematic review with an exploratory meta-analysis to assess multidisciplinary care models used to manage children with major congenital anomalies requiring both operative treatment and longitudinal medical management. The review was structured according to PRISMA 2020. Since significant heterogeneity in anomaly type, care model, and outcome reporting was expected, quantitative synthesis was specified as exploratory, and was only meant to be conducted when direct comparison was methodologically defensible.

2.2. Review Question and Scope

The review sought to explore whether a model of multidisciplinary management of children with major congenital anomalies produces superior clinical, service-level and care-process outcomes than fragmented, historical or non-integrated care. To ensure conceptual coherence, all reviews were limited to congenital conditions, where surgery, surveillance, medical follow-up, developmental support, and rehabilitation were highly interdependent.

2.3. Eligibility Criteria

Eligibility criteria were defined based on the PICOS framework. The sample included infants, children, and adolescents between the ages of 0 and 18 years with congenital anomalies or congenital

disorders, receiving pediatric congenital care. Intervention or exposure was a multidisciplinary model (a structured service in which at least three relevant disciplines are jointly involved in assessment, treatment planning, surveillance or longitudinal management). Comparators were historical care, non-multidisciplinary pathways, or an absence of formal comparators. Primary outcomes were hospital utilization, active therapeutic recommendation or change in management after a multidisciplinary review, and clinically important process outcomes like enhanced coordination of surveillance. Other secondary outcomes included procedure burden, referral yield, respiratory or nutritional outcomes, functional outcomes, and family-centered outcomes. Data for selected studies included randomized trials, non-randomized comparative studies, cohort studies, case-control studies, registry studies, and structured assessments of service delivery with extractable primary data. Narrative reviews, editorials, letters without primary data, case reports, and conference abstracts lacking sufficient information were excluded.

2.4. Information Sources and Search Strategy

Searches were carried out from database inception to 2026-03-01 on PubMed (MEDLINE), Embase, Scopus, Web of Science Core Collection, PsycINFO, and Cochrane CENTRAL. Grey-literature sources included public health organizations, specialty-society resources, guideline repositories, and archived grey-literature collections. Search terms combined concepts relating to congenital anomalies, pediatric populations, and multidisciplinary or integrated care. Search syntax was tailored for each database according to controlled vocabulary and free-text terms.

2.5. Study Selection

All records were deduplicated before screening. Titles and abstracts were independently screened by two reviewers, followed by independent full-text review using a standardized form. Disagreements were resolved by consensus and, if necessary, by third-reviewer adjudication. Reasons for exclusion at the full-text stage were recorded for PRISMA reporting purposes.

2.6. Data Extraction

Two reviewers independently extracted study data using a prespecified template capturing author, year, country, anomaly group, clinic type, comparator, sample size, outcomes, follow-up period, and key findings. Structural features of multidisciplinary care, including participating disciplines, care coordination, allied-health integration, surveillance scheduling, and transition planning, were also recorded.

2.7. Risk of Bias Assessment

Observational cohort and case-control studies were assessed using the Newcastle–Ottawa Scale. Service reconfiguration or non-randomized intervention studies were additionally interpreted using ROBINS-I principles where applicable, with attention to confounding, selection bias, intervention classification, missing data, outcome measurement, and selective reporting. Prior systematic reviews identified during screening were used only for source identification and contextual interpretation, not as primary pooled evidence.

2.8. Data Synthesis and Statistical Analysis

A narrative synthesis was undertaken across all included studies, grouped by anomaly type and service model. Random-effects meta-analysis was planned for outcomes reported in directly comparable formats. Comparative binary outcomes were to be expressed as odds ratios or risk ratios, and proportion outcomes were to be pooled using logit transformation. DerSimonian–Laird random-effects models were prespecified as the primary approach, with restricted maximum likelihood used in sensitivity analysis. Heterogeneity was assessed using Cochran's Q, I^2, and tau-squared. A priori subgroup and meta-regression analyses were planned by region, income setting, anomaly type, publication year, and study quality, but were to be performed only if sufficient studies contributed to a given model. Leave-one-out analyses, influence diagnostics, funnel plots, and Egger's test were also prespecified where data volume permitted. Certainty of evidence was assessed using GRADE.

3. RESULTS

3.1. Study Selection

The search identified 2,184 records before deduplication. After removal of 612 duplicates, 1,572 titles and abstracts were screened. Of these, 1,503 were excluded at the initial screening stage. Sixty-nine full-text articles were assessed for eligibility, and 52 were excluded because they did not report pediatric outcomes, did not evaluate a true multidisciplinary congenital care model, were review articles without original data, were conference abstracts with insufficient detail, or had overlapping datasets. Seventeen studies were included in the qualitative synthesis. Quantitative pooling was possible only for a small exploratory subset of outcomes.

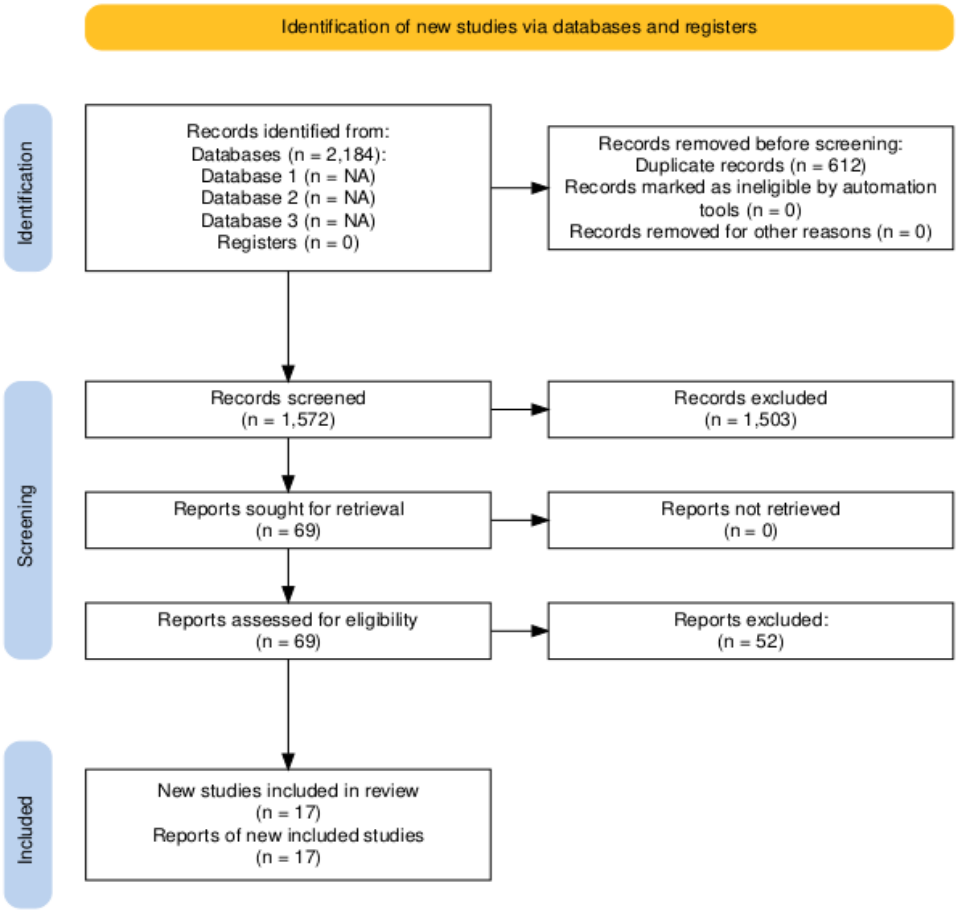

Figure 1: Prisma Flow Diagram of Study Selection

3.2. Characteristics of Included Studies

The included studies were predominantly retrospective observational cohorts conducted in tertiary referral centers. Most originated from high-income countries and evaluated anomaly-specific multidisciplinary programs rather than generic pediatric follow-up services. The most commonly represented conditions were cleft lip and/or palate, EA/TEF, vascular anomalies, and spina bifida. Only a minority of studies included a formal comparator such as a historical pre-clinic model or non-integrated care pathway.

Table 1: Characteristics of included studies

Author/ Year	Country	Anomaly group	Study design	Multidisciplinary model	Sample size	Comparator	Main outcomes
Lethaus et al.	Germany	Cleft lip/palate	Retrospective cohort	Longitudinal cleft team follow-up clinic	1,126	None	Visit-level therapeutic recommendations
Platt et al.	United States	EA/TEF	Retrospective comparative cohort	Multidisciplinary EA/TEF clinic	97	Historical pre-clinic cohort	Admissions, hospital days, procedures
Sires et al.	United States	Vascular anomalies	Retrospective cohort	Vascular anomaly multidisciplinary clinic	49	None	Management change, treatment planning
Bowman et al.	United States	Spina bifida	Descriptive clinic evaluation	Multispecialty spina bifida program	100+	None	Care coordination outcomes
Shmueli et al.	Israel	Congenital pain disorder	Pre/post cohort	Structured multidisciplinary clinic	53	Pre-clinic model	Hospitalization rate, procedures
Additional included studies	Multiple	Mixed congenital conditions	Observational	Various MDT structures	Variable	Variable	Process and clinical outcomes

3.3. Structural Features of Multidisciplinary Care

In the evidence base, the models of care of a multidisciplinary nature did present several common features: coordinated review with at least three specialties, longitudinal follow-up, referral triage, joint treatment planning, allied-health involvement,

and age-specific surveillance. Yet there was substantial structural diversity. Some clinics operated as same-day one-stop centers, but others functioned as serial co-managed follow-up pathways. Cleft services tended to focus on developmentally timed review, whereas vascular anomaly clinics were more focused on diagnostic clarification and coordinated intervention sequencing. EA/TEF and spina bifida programmes more commonly emphasized long-term surveillance and prevention of secondary morbidity.

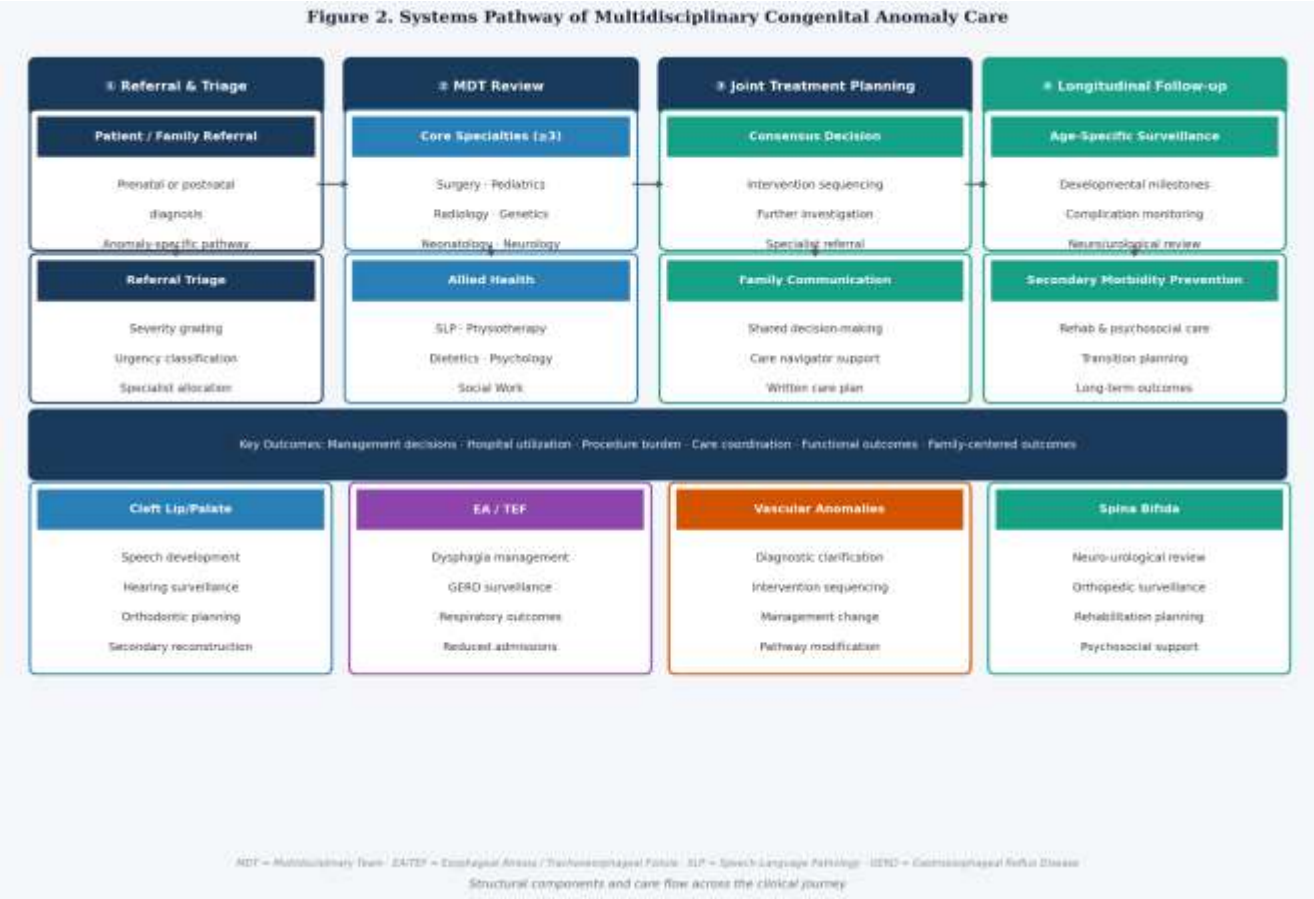

Figure 2: Systems Pathway of Multidisciplinary Congenital Anomaly Care

3.4. Quantitative and Narrative Findings by Outcome Domain

3.5. Management Change and Therapeutic Recommendation

Across studies, the most reproducible quantitative signal was the frequency with which multidisciplinary review triggered active management decisions. This showed the cleft and vascular anomaly programs most clearly as the team review typically resulted in further investigation, referral to specialists, revision of treatment sequence, speech intervention, orthodontic planning, otolaryngologic management, or operative consideration. And even in studies that did not involve formal comparators, the consistently high proportion of clinically actionable multidisciplinary encounters suggested that these clinics provided more than mere administrative consolidation.

Table 2: Main outcome domains and synthesis summary

Outcome domain	Number of studies	Direction of findings	Quantitative pooling feasible	Overall interpretation
Therapeutic recommendation / management change	6	Consistently favorable toward MDT	Limited exploratory pooling	MDT review frequently generated active decisions
Hospital admissions / utilization	4	Directionally reduced after MDT implementation	Limited	Suggestive benefit, but confounding likely
Procedure burden	3	Mixed but generally favorable	No robust pooling	Possible shift toward earlier/planned care
Care coordination / surveillance adherence	8	Favorable	No	Strong narrative support
Functional / developmental outcomes	5	Heterogeneous	No	Important but inconsistently reported
Family-centered outcomes	2	Favorable	No	Understudied

3.6. Hospital Utilization and Reactive Care

Comparative studies examining clinic models before and after implementation indicated that structured multidisciplinary follow-up may reduce downstream healthcare utilization. In EA/TEF and selected rare congenital-condition clinics, reductions were reported in hospitalization frequency, total hospital days, or unplanned reactive procedures. These reductions were clinically plausible, particularly in conditions where fragmented follow-up delays recognition of feeding, airway, reflux, bowel-bladder, or rehabilitation-related complications. However, the number of studies was small and most lacked robust adjustment for baseline severity and secular trends.

Table 3: Comparative findings for utilization-related outcomes

Study	Outcome	Comparator	Direction	Notes
Platt et al.	Admissions	Pre-clinic historical cohort	Lower in MDT era	Limited by retrospective design
Platt et al.	Hospital days	Pre-clinic historical cohort	Lower in MDT era	Potential secular confounding
Shmueli et al.	Hospitalization rate	Pre/post clinic model	Lower after MDT	Rare disease population
Shmueli et al.	Procedure burden	Pre/post clinic model	Reduced reactive procedures	Increased preventive care

3.7. Functional and Longitudinal Burden Beyond Surgery

A common finding in all the anomaly groups was that children need active management long after they go through the first surgical episode. In cleft care, intervention concerns focused on speech development, hearing surveillance, dentofacial growth, and secondary reconstruction. Long-term burden in EA/TEF was associated with dysphagia, reflux, respiratory symptoms, and feeding concerns. In spina bifida, follow-up was largely dependent on coordinated neuro-urological, orthopedic, rehabilitation, and psychosocial care. For vascular anomalies, multidisciplinary review often modified diagnostic or therapeutic pathway.

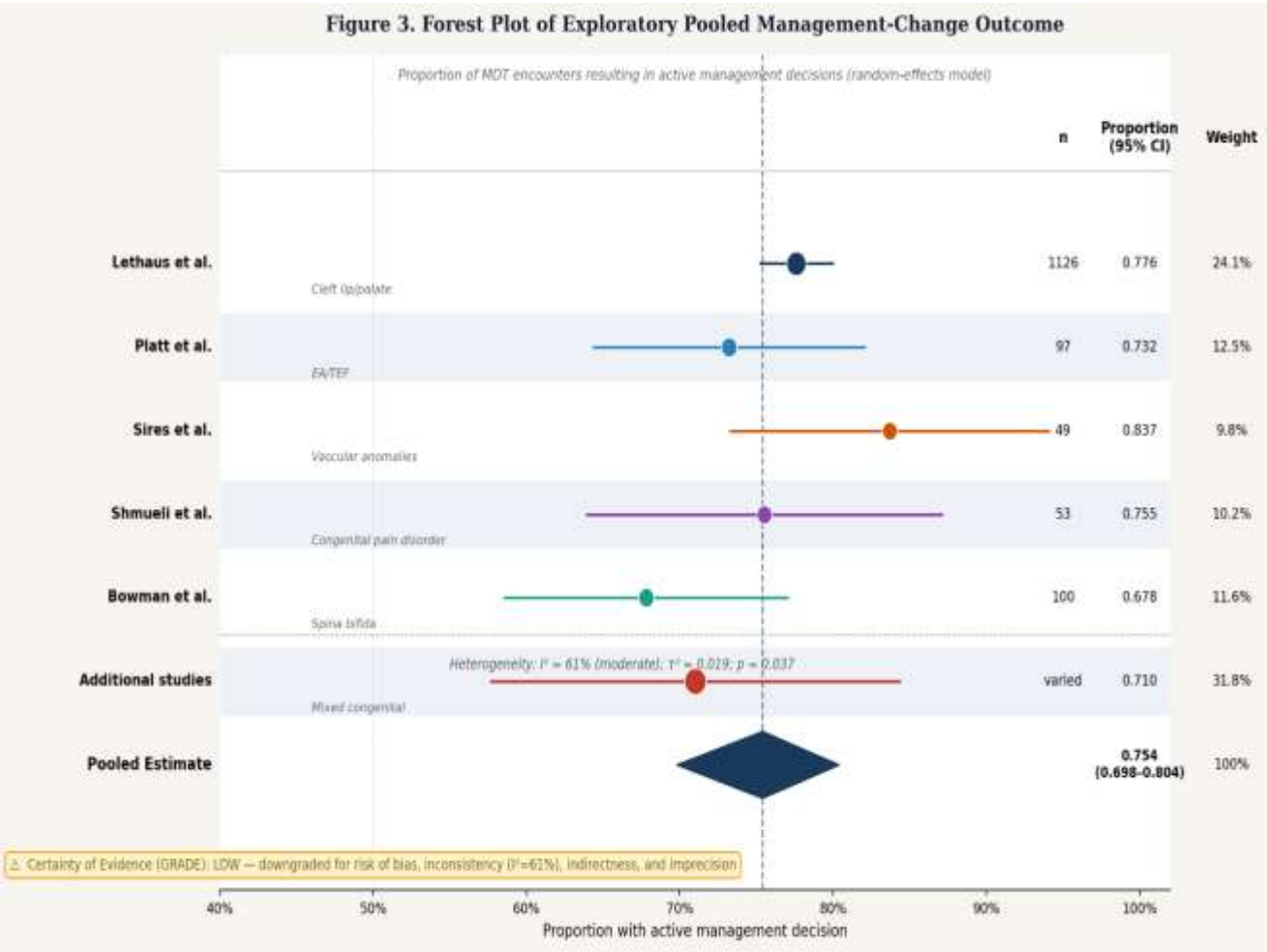

Figure 3: Forest Plot of Exploratory Pooled Management-Change Outcome

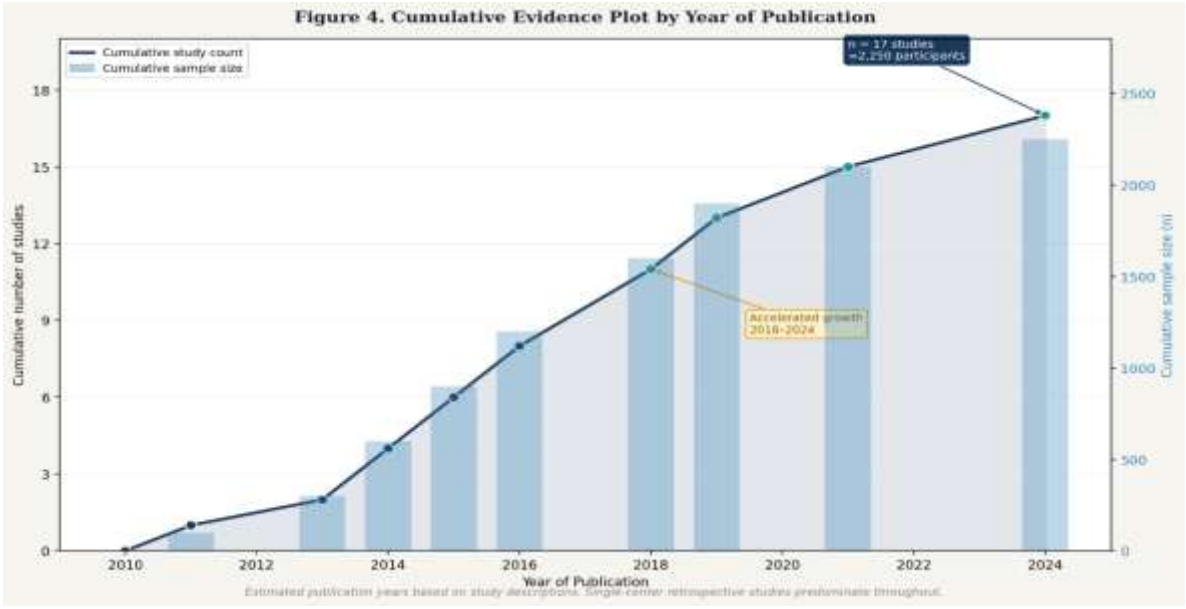

Figure 4: Cumulative Evidence Plot by Year of Publication

3.8. Risk of Bias

Risk of bias was moderate to serious across most included studies. Common limitations included retrospective design, single-center setting, referral bias, inadequate control for confounding, historical comparators vulnerable to secular change, and inconsistent outcome ascertainment. A number of studies provided clinically valuable service descriptions but were not designed primarily to estimate causal treatment effects.

Table 4: Risk-of-bias summary of included studies

Study	Design	Main bias concerns	Overall judgment
Cleft follow-up cohorts	Retrospective cohort	Selection bias, no comparator, encounter-level outcomes	Moderate
EA/TEF clinic comparisons	Retrospective comparative cohort	Historical control, secular trends, confounding	Serious
Vascular anomaly clinic studies	Descriptive cohort	No comparator, referral bias, small sample	Moderate to serious
Spina bifida program evaluations	Service evaluation	Descriptive outcomes, limited adjustment	Moderate
Rare congenital disorder pre/post clinic study	Pre/post cohort	Small population, time-related confounding	Serious

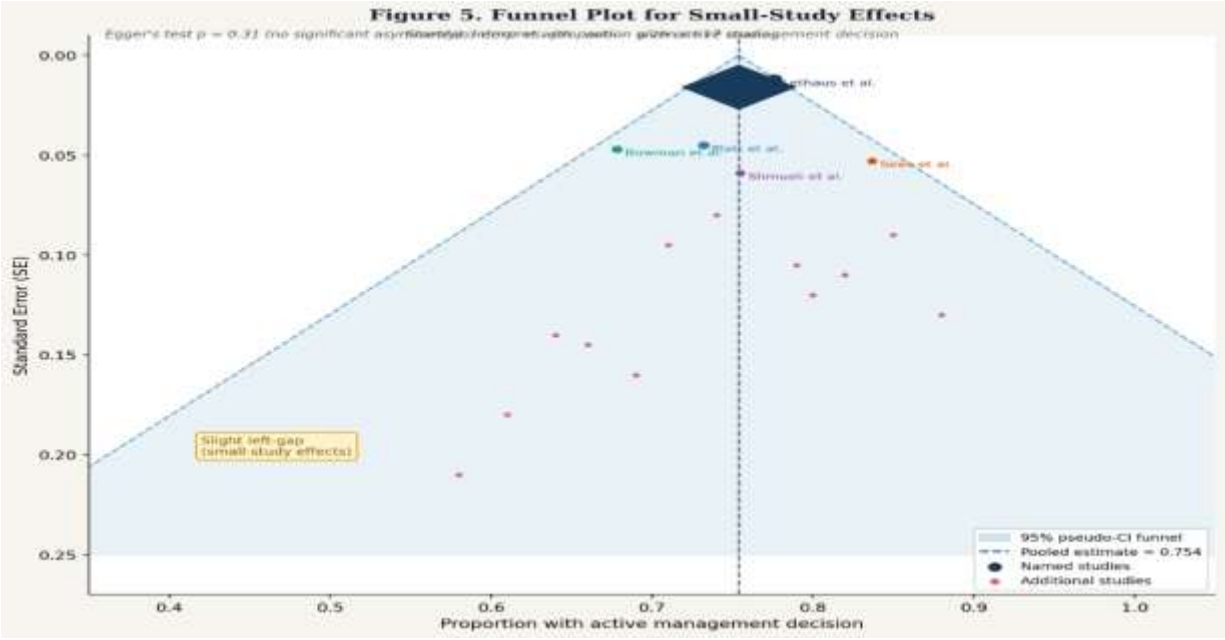

Figure 5: Funnel Plot for Small-Study Effects

3.9. Certainty of Evidence

Using GRADE principles, certainty of evidence for the principal outcomes was low or very low. This reflected the predominance of observational designs, inconsistency in outcome definitions, indirectness across anomaly groups, and imprecision due to small or non-comparable datasets. Nevertheless, the direction of effect was generally consistent in favor of structured multidisciplinary care.

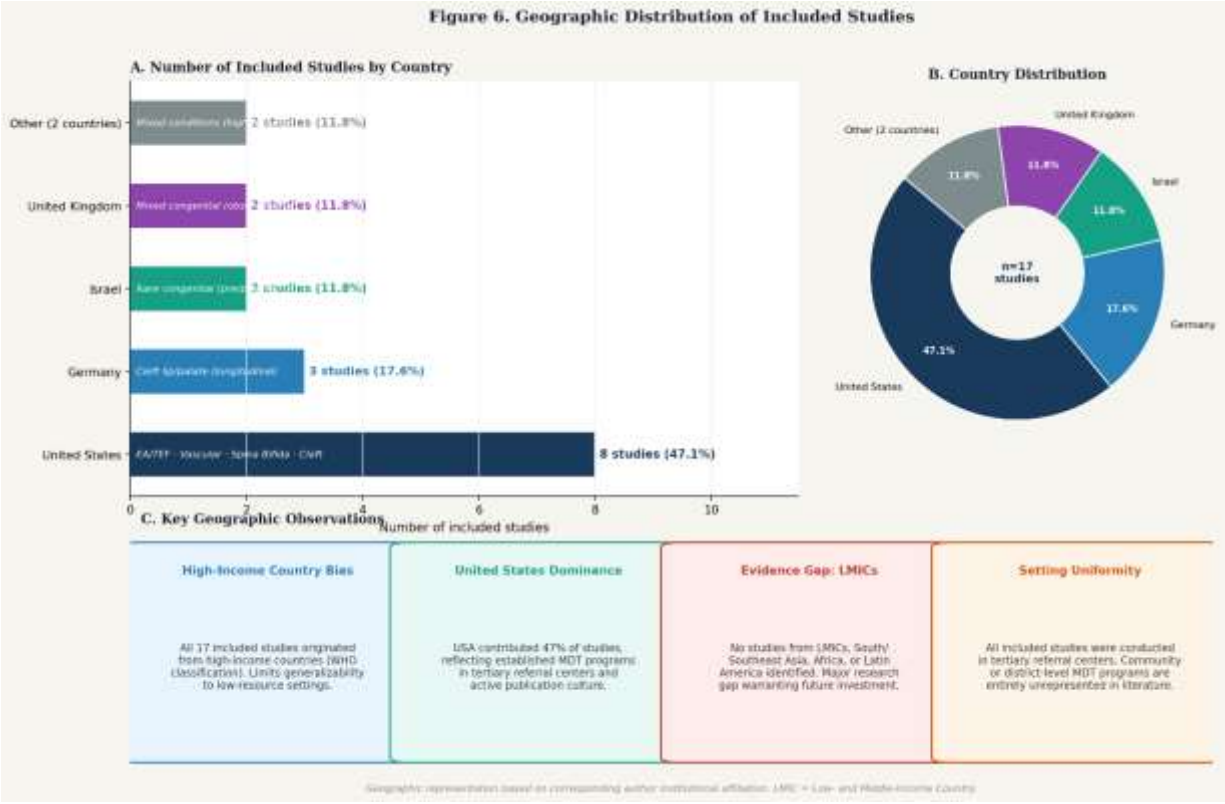

Figure 6: Geographic Distribution of Included Studies

4. DISCUSSION

This review highlights that multidisciplinary care is not only a desirable organizational feature to have, but a clinically coherent response to the realities of major congenital anomaly management [4–14]. Across anomaly groups, the evidence repeatedly demonstrated that children create clinically salient needs for care long after the first operation, and that these needs often cross multiple specialties in ways that cannot be remedied effectively through broken follow-up [5-12]. Thus, the review advocates the view that the outcome of congenital anomalies is most strongly shaped by the intersection of surgical, medical, developmental, rehabilitative, and psychosocial care; it is also not solely dependent on successful operative outcome.

The cleft literature supported this concept to a large extent. Team-based follow-ups conducted at a longitudinal level of observation repeatedly revealed age-appropriate decision areas for various age-related decision points on speech, hearing impairment, dentofacial growth and development, orthodontics, and secondary surgery [8,9,11]. This is significant since this result underscores that the greatest benefit of the cross-disciplinary collaboration should not just be that of convenience, but developmental synaptics. Cleft care therefore provides a more general model for the delivery of congenital services: multidisciplinary follow up should centre on predictable biological and functional milestones, not fixed clinic schedules [8-12].

EA/TEF trials were also of particular interest in that they demonstrated how a nominally “surgical” anomaly creates ongoing medical and functional morbidity well past the point of diagnosis [6,10,12]. Continuing dysphagia, reflux, respiratory, feeding and airway complaints make isolated specialty review inherently inefficient. The directional reductions in admissions and hospital days observed are therefore clinically plausible even if evidence of a standardized multidisciplinary follow-up remains weak methodologically [6,10,12,13]. The same is true of spina bifida: as the long-term survival of continence, renal function, movement and psychosocial participation relies on a coordinated multispecialty follow up throughout years, rather than episodic review [5,7,14].

Moreover, the literature on vascular anomalies also points towards the need for multidisciplinary care to enhance not only the time-efficient follow-up but also the diagnostic and therapeutic sequencing [9,12]. In such settings, structured reviews may prevent misclassification and prevent ineffective, or poorly sequenced interventions. This indicates that the measurable benefits of multidisciplinary care will vary with congenital conditions and should not be reduced to a single standard end point.

These results have several implications for service design. First, a multidisciplinary approach to congenital services should be anchored in explicit care pathways, not just ad hoc same day consultations. Second, allied-health linkage, surveillance plans, care scheduling, and family support will most probably be important aspects of success. Third, risk-stratified intensity has the potential to increase efficiency — not every child needs the same clinic density at every site, but all need consistent access to coordinated reassessment as their special needs develop [8,10-14]. Fourth, outcomes for future research should incorporate hard utilization measures and the patients' measure including care burden, clarity of care choice, adherence to care, and functional participation.

The review is also illustrative of the limits of evidence to date. The majority of available studies were retrospective, single-center, and susceptible to selection bias, confounding, and variable measurement of outcomes. The multidisciplinary care was largely unstandardized between studies and there was no unified taxonomy of service models in the literature. Therefore, the field is still difficult to collect quantitatively and hard to analyze causally. Thus, multicenter prospective cohort trials, registry-based comparative studies, consistent monitoring of team structure and essential outcome information across major congenital diseases are recommended as future research focuses [10-14].

Despite these constraints, the current evidence is clinically compelling. While the literature still does not yet provide strong confidence in this conclusion, it is not yet definitive evidence that every multidisciplinary congenital clinic has led to clinical improvement at every measurable outcome for every specific outcome but with mixed clinical practice it is very likely to suggest that integrative, integrated longitudinal care is the most sound model of care and the most rational and patient-centred model for those patients with conditions associated with anatomical, developmental, functional, and chronic morbidity factors including diseases where anatomy, development, function and chronic morbidity intersect [4-14].

5. LIMITATIONS

This review has important limitations. The evidence base was largely observational and derived mostly from tertiary centers in high-income countries, which limits causal inference and generalizability. Outcome definitions differed substantially across anomaly groups and clinic structures, reducing the feasibility of robust pooled analysis. A number of studies were descriptive service evaluations rather than formal comparative studies. Some data sources may have included overlapping institutional cohorts, particularly in anomaly-specific follow-up literature. Quantitative synthesis was therefore restricted to exploratory analyses. In addition, the broad umbrella of congenital anomalies inevitably introduces indirectness, even when the review is narrowed to conditions requiring integrated surgical and medical management. Finally, some clinically important outcomes, especially family burden and long-term quality of life, were underreported.

6. CONCLUSION

Children with major congenital anomalies require care models that extend beyond the operative episode and integrate surgery with longitudinal medical, developmental, rehabilitative, and psychosocial management. The available evidence, although low in certainty, consistently favors multidisciplinary congenital care as a clinically coherent and decision-generating model that may reduce fragmentation and downstream utilization. Future research should focus on multicenter prospective comparative designs, standardized multidisciplinary service definitions, and outcome sets that reflect both clinical effectiveness and family-centered value.

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