

How to advise women with a medical history of surgically corrected anorectal malformation on vaginal delivery: a systematic review

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Abstract

Objective: Discussion remains on how to advise women with a past medical history of surgically corrected anorectal malformations (ARMs) regarding vaginal delivery. The aim of this review is to evaluate and review the reported obstetrical complications and outcomes after vaginal delivery for these women.

Data sources: A systematic search was performed from inception up to 8 December 2020 in PubMed, Embase.com and Clarivate Analytics/Web of Science Core Collection, with backward citation tracking.

Study eligibility criteria / appraisal: All articles reported on the outcomes of interest in women with a past medical history of surgically corrected anorectal malformation and had a vaginal delivery were included with the exception of editorial comments or invitational commentaries. Screening and data extraction was done by two authors independently with a third and fourth reviewer in case of disagreement. Tool for Quality assessment depended on the type of article. As low quality evidence was expected no meta-analysis was performed.

Results: Only four of the 2125 articles screened were eligible for inclusion with a total of 12 vaginal deliveries attempts in 7 women. In two patients complications were reported: failed vaginal delivery requiring urgent cesarean section (n=1), vaginal tears (n=1).

Conclusion: high quality evidence regarding outcomes and complications after vaginal delivery in women with a history of surgically corrected anorectal malformation is lacking. Therefore, based upon this systematic review no formal recommendation can be formulated regarding its safety. Future studies are essential to address this problem.

Prospero registration: CRD42020201390. Date: 28-07-2020s

1. Background

Anorectal malformations (ARMs) are rare congenital malformations with an estimated incidence of approximately 1 in 5000 live births per year in the Western civilization.^{1,2} The diagnosis is usually made in the early neonatal period and affects both male and female equally.³ As known, a wide variety of anorectal malformations exist, ranging from perineal fistulas to more complex fistulas, even to the urogenital tract. In general, functional outcome is worse in patients with more complex types of anorectal malformation, but it also depends on other factors such as the presence of other associated anomalies (spine, spinal cord and urogenital anomalies). In females, types of anorectal malformation encountered are isolated imperforate anus (4.8%), rectovestibular fistula (60.3%), rectoperineal fistula (20.6%) and cloacal anomalies (7.9%).^{1,2,4,5} Cloacal malformations can be subdivided based on the length of common channel, namely < 3 cm and > 3 cm.³ In most cases of anorectal malformation, surgical correction is necessary, usually at an early age.

As mentioned above, female patients with anorectal malformation may also suffer from other disease-specific problems related to the gynecological tract as anatomical anomalies, sexual/intercourse problems, fertility and obstetrical complications.⁶ In up to 20% of patients with rectovestibular fistula, gynecological

abnormalities, such as a vaginal septum, bicornuate uterus or in some rare cases even a vaginal agenesis.⁴ Not only the disease (anorectal malformation and its associated anomalies) itself, but also the surgery needed to correct the anomalies may affect the gynecological tract. This in turn has consequences later on in life in female patients with a medical history of surgically corrected anorectal malformation. For example, increased damage can be expected during vaginal delivery in a scarred reconstructed perineum. The anal sphincter and in some women reconstructed urethra or vagina, may be at risk for dysfunctioning after vaginal delivery, caused from the significant stretch or even ruptures of the perineum.⁷ Intensive guidance and counselling regarding pregnancy and mode of delivery in patients with anorectal malformation is therefore essential. It is generally recommended to perform a cesarean section (CS) for all patients with a past medical history of anorectal malformation. Some surgeons however believe that in certain types of anorectal malformation, for instance rectovestibular or rectoperineal fistulas, vaginal delivery could be possible.² To our knowledge evidence regarding this topic is scarce, regardless of type of anorectal malformation. As a result, current recommendations seem to be based on low-quality evidence (expert opinions).

2. Objective

Therefore, the aim of this systematic review is to determine which obstetrical complications and outcomes have been reported for women with a past medical history of surgically corrected anorectal malformation that gave birth vaginally.

3. Materials And Methods

The protocol of this systematic review was registered at PROSPERO: International prospective register of systematic reviews with identification number CRD42020201390. This systematic review was reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement.⁸ Although this systematic review was performed due to questions from our patients, they were not actively involved in the design and conduct of this review. Nor did this research receive any funding.

a. Eligibility criteria, information sources, search strategy.

A systematic search was performed (by AW and JK (medical librarian)) from inception up to 8 December 2020 in PubMed, Embase.com and Clarivate Analytics/Web of Science Core Collection. Keywords (including synonyms and closely related words) were anorectal malformations, cloaca, imperforate anus, natural or vaginal delivery. The complete search strategy is shown in Appendix A. To ensure that all possible publications were included, the citations list from all full text screened articles were checked. Only articles that reported our primary outcome, namely the number of patients with complications after vaginal delivery in women with a past medical history of surgically corrected anorectal malformation, were included in this systematic review. All types of studies are included, with the exception of editorial comments and invited commentaries. Language was restricted to English and Dutch. Articles describing our primary outcome in women with Hirschsprung disease and women with a sphincter rupture in their past medical history (without anorectal malformation) were excluded from this study. In addition, studies describing only the outcomes of

cesarean section were also excluded. There were no restrictions in age or type of anorectal malformation. The primary outcome was the number of patients with complications after vaginal delivery in women with a past medical history of surgically corrected anorectal malformation, as reported by the original paper. Severity of complications were assessed according to the Clavien-Dindo Scale.^{9,10} Secondary outcomes were the type of perineal- and sphincter ruptures as reported by the original paper. Also, other obstetrical outcomes such as defecation problems, sexual problems and urinary problems as reported by the original paper were collected. To our knowledge a core outcome set for this specific patient group is lacking.

b. Study selection

Abstracts were screened independently by two reviewers (AW, TA) according to the in- and exclusion criteria as described above. After which, full text assessment of the selected articles was performed. Disagreements were resolved by consulting a third and fourth reviewer (RG, JD). If studies seemed eligible, but individual data or the primary outcome was lacking/not identified in the study, the first and last author were contacted by mail to obtain more information about their article in order to include them in our systematic review.

c. Data extraction

After full-text screening, data extraction was done by two reviewers independently (AW, TA) using a standardized data extraction form. Again, disagreements were resolved by a third and fourth reviewer (RG, JD).

d. Assessment of risk of bias

Risk of bias was assessed using the appropriate tool according to the type of study, for instance for RCTs we planned to use the Cochrane risk of bias tool.¹¹ For comparative cohort studies, we anticipated to use the Newcastle-Ottawa scale if possible.¹²

e. Data synthesis

As it was expected to encounter a low number of studies of low quality it is already anticipated to not perform a meta-analysis. Instead only descriptive variables of the included studies will be displayed. Regarding our primary outcome the number of patients will be displayed. Results are presented in various tables and figures, as absolute numbers.

4. Results

a. Study selection

The search yielded a total of 4339 articles. After removal of duplicates, 1892 articles were screened by title and abstract. In addition, we screened 233 articles through the citations lists from the concerning articles. In total, 2125 articles were screened. Overall, out of the 49 articles that were assessed by full-text, four articles were eligible for inclusion (Fig. 1).

b. Study Characteristics

a. Included studies

The general characteristics of the included studies are shown in Table 1.¹³⁻¹⁶ In the two retrospective case series, complications during vaginal delivery were not specifically mentioned.^{13,14} Because of this, contact was made, as mentioned above, Pena et al. reported that no complications had occurred.¹⁴ Unfortunately, we did not receive an answer from Iwai et al. in the article they stated that all women had a normal vaginal delivery.¹³ Therefore, the assumption was made that these women did not experience any complications during delivery. For this reason, both articles were included. Appiah-Sakyi reported a woman who failed to deliver vaginally after an hour of pushing, after which a cesarean section was performed.¹⁵ They discovered that she had a blind-ending pouch of her uterine cavity which had no connection with the cervical canal. Because of missing data regarding the anatomical description and how she got pregnant, contact was made with the authors.¹⁵ Unfortunately, we did not receive an answer. Thereby the assumption was made that she had a non-communicating rudimentary horn with cavum, in which a vaginal delivery was never an option.

Furthermore, there is a large heterogeneity among the articles due to the missing data.

b. Excluded studies

From the 49 articles who were screened full-text, 45 articles were excluded. Reasons for exclusion can be found in Fig. 1 and appendix B. Three articles require additional explanation as there was disagreement between the first two reviewers requiring assessment by the third and fourth reviewer.

Vilanova-Sanchez et al., a systematic review, included 13 articles about obstetrical outcomes in women with a past medical history of an anorectal malformation.⁶ In total 24 pregnancies were reported.⁶ Two articles included both one patient with a vaginal delivery.^{6,16,17} One was already included in our review, the other one did not meet the criteria of surgery in the past medical history.^{16,17}

Greenberg et al. 2003, a case report, describes the same patient described in the article of 1997 which was included in our review.^{16,18} This second report describes her second pregnancy, after which she delivered through a cesarean section.¹⁸

Finally, contact was made with the authors of Davies et al. because of lacking information.¹⁹ Unfortunately, the needed information was not available, so no assumptions could be made. Therefore, the article was excluded.

c. Risk of bias of included studies.

In this review, two case reports and two case series were included.¹³⁻¹⁶ As the original New-Castle Ottawa scale is not applicable for case series, it was decided to use the adapted version as described by Hassan Murad et al.²⁰ The two case series scored poorly, focusing on selection bias and poor description of method. Looking at all the evidence we have gathered, we can state that level four (poor quality) evidence according to the Oxford Centre for Evidence-Based Medicine for our primary outcome is present.²¹

d. Synthesis of results

Baseline characteristics of the included studies are displayed in Table 1.¹³⁻¹⁶ Gestational age of the babies was only reported by Greenberg et al. and Appiah-Sakyi et al. and were respectively 34.5 weeks and 32 weeks.^{15,16} Greenberg et al. reported also type of pain relief and birth weight, namely an epidural and 2310 gram.¹⁶

Table 1
General characteristics of the included studies.¹²⁻¹⁵

	Study design	Number of patients	Type of ARM	Type of correction	Age, years	OB history	Type of delivery
Greenberg et al. 1997	Case report	1	Cloaca	Colostoma, Rectal pull through procedure, repair of rectovaginal fistula	27	G1P0	Vaginal VAVD FAVD
Iwai et al. 2007	Retrospective case series	3	#1. High type ARM #2. Low type ARM #3. Low type ARM	#1. Colostomy + abdominoperineal rectoplasty #2 and #3. Neonatal perineoplasty	#1. 31 #2. 40 #3. 25	#1. G? P2 #2. G? P5 #3. G? P1	Vaginal
Pena et al. 2004	Retrospective case series	2	Cloaca with common channel < 3cm	Posterior sagittal approach with total urogenital mobilization (TUM)	N/R	N/R	Vaginal
Appiah-Sakyi et al. 2009	Case report	1	Imperforate anus	N/R	25	G1P0	CS
OB history, obstetrical history; G (gravida) P (para); N/R, not reported in original article; VAVD, vacuum-assisted vaginal delivery; FAVD, forceps-assisted vaginal delivery, CS; cesarean section.							

I. Primary outcome

Our primary outcome is displayed in Table 2. In total we included in this systematic review 12 vaginal deliveries attempts in 7 women. Of these attempts, one patient required a cesarean section as vaginal delivery failed. Due to the lack of information on the anesthesia techniques, this complication was scaled as Clavien-Dindo IIIA/IIIB.¹⁵ In another woman two tears (one large left vaginal sulcus tear (Grade IIIA) and small midline introital tear (Grade IIIA)) occurred after Tucker-McLean Forceps were applied requiring repair.¹⁶ Therefore complications occurred in 2 women.

Table 2
Overview primary outcome in the included studies.¹²⁻¹⁵

	Number of patients	Total number of vaginal deliveries	Total number of complications	CD classification
Greenberg et al. 1997	1	1	2	Grade IIIA
Iwai et al. 2007	3	8	0	N/A
Pena et al. 2004	2	2	0	N/A
Appiah-Sakyi et al. 2009	1	0	1	Grade IIIA or Grade IIIB
N/A, not applicable; N/R, not reported.				

II. Secondary outcomes

Table 3 displays the secondary outcomes for each study. Functional outcome was reported in only two studies.^{15,16} Greenberg et al. reported no defecation problems and Appiah et al. reported no urinary or fecal incontinence.^{15,16} However, it is unclear what kind of follow-up was done. Sexual problems were not discussed in the articles.

Table 3
Overview of the specific obstetrical complications in the included studies.¹³⁻¹⁶

	Complications	CD classification	Functional outcome
Greenberg et al. 1997	#1. Large left vaginal sulcus tear #2. Small midline introital tear	#1. Grade IIIA #2. Grade IIIA	Well healed. Complete return of baseline bowel function
Iwai et al. 2007	No complications	N/A	N/R
Pena et al. 2004	No complications	N/A	N/R
Appiah-Sakyi et al. 2009	CS	Grade IIIA or Grade IIIB	No residual urinary or fecal incontinence.
N/A; not applicable. N/R; not reported in original article. CS; cesarean section.			

5. Discussion

Principal findings

This systematic review shows that literature regarding obstetrical outcomes and complications after vaginal delivery in women with a medical history of surgically corrected anorectal malformation is scarce and of low

quality. Formal recommendation on the mode of delivery can therefore not be made. Paucity of the literature indicates the necessity of large studies investigating the obstetrical outcomes and complications in women with surgically corrected anorectal malformation.

Comparison with existing literature

To our knowledge, this is the first systematic review according to the PRISMA methodology performed on this subject, providing new information that can be used to counsel pregnant patients with a medical history of surgically corrected anorectal malformation. A recent literature study on this matter concluded that a cesarean section is preferable in patients with a cloacal repair, as these patients all have undergone some type of extensive correction of the perineal body and vagina.⁶ These patients would have an increased risk of damage to these structures during vaginal delivery because scar tissue does not stretch as well as healthy tissue. The authors based their conclusion on their review of 13 articles of which only two articles reported results of vaginal delivery.⁶ In line with our finding, they also stated that there is a paucity of evidence-based data.

In our systematic review, only four articles could be included, which all were of low-quality due to the study design with consequent methodological flaws. As our interest was vaginal delivery, we did not report the outcomes after a cesarean section which led to the exclusion of 18 articles. Only Appiah et al. was included because vaginal delivery was tried before performing a cesarean section.¹⁵ This study shows the importance of screening for gynecological malformations in female patients with anorectal malformation as anatomical abnormalities might be present, that may affect the choice of delivery method.¹⁵

Most clinicians consider cloacal malformations as complex anorectal malformations requiring extensive surgery at young age. These patients might be prone to damage to birth canal and pelvic floor by extensive stretching during vaginal delivery that may result in ruptures. Therefore, in most patients, a cesarean section is advised. In other anorectal malformations recommendations are not specifically made. Another possible reason why there are relatively more cesarean sections may be due to cultural differences. More cesarean sections are conducted in for example Latin America and the Caribbean region.²²

The data on which the advice to do a cesarean section is made is lacking in the current literature. One must bear in mind that a cesarean section might also be a potential harmful procedure for both mother and child. For example, the incidence of postoperative ileus after cesarean section lies around the 12%.²³ In addition, there is a higher risk of postpartum sepsis, especially in an emergency cesarean section, and women who deliver by cesarean section are more likely to be admitted to the ICU.^{23,24} Subsequent cesarean section carry even higher risks. Kramer et al. found a 47% increase in abnormal placentation and 40% increase in placental abruption.²⁴ The number placenta accretes correlate with the number of cesarean section in the past medical history.²⁴ For 1–5 cesarean sections in the past medical history the percentages are respectively 3%, 11%, 40%, 61% and 67.1%.²⁴ Furthermore, some women, especially the ones with a cloaca, have undergone additional procedures like a bladder augmentation.⁶ These procedures warrants caution when performing a cesarean section due to the risk of iatrogenic damage. However, the risk of bleeding in a planned cesarean section is lower in comparison to a planned vaginal birth (respectively 1.1% and 6.0%).²³

Apart from adverse effects on the mother, a cesarean section can have disadvantages for the child as well. The lungs of a newborn should be cleared rapidly to allow gas exchange for a smooth transition to air breathing.²⁵ The lung is full of fluid and should be cleared rapidly to allow gas exchange.²⁵ If not it can result in respiratory morbidity.²⁵ Tefera et al. performed a systematic review and meta-analysis on the risk of neonatal respiratory morbidity in elective cesarean section vs vaginal delivery.²⁵ They found more respiratory problems in children born by elective cesarean section versus vaginal delivery.²⁵

The discussion about the risks in mother and child continues. A careful consideration must be made looking at both mother and child. This issue deserves further international attention.

Around 2.4% of the women who deliver vaginally develop obstetrical anal sphincter injuries (OASI).²⁶ Studies show that tears of the perineum are risk factors for developing urinary incontinence, fecal incontinence and dyspareunia.^{26,27} Because of these risks, vaginal deliveries can have disadvantages as well. Additional research is needed to provide a recommendation in anorectal malformation patients.

Strengths and limitations

This systematic review only included four studies of poor quality mainly due to the methodology used (i.e. case series). Large heterogeneity in these series therefore existed, not only regarding patient selection, but also outcome definitions. Although in most studies general statements regarding complications of vaginal delivery were made, only two study explicitly described them.^{15,16} Moreover, secondary outcomes that we identified were absent in most of these studies and therefore no conclusions could be drawn. Secondly due to the small sample size and poor-quality data, no general recommendation can be made. Thirdly as mentioned above selection bias is present as in most studies it is unclear why patients gave vaginal birth. There is also selection bias/indication bias due to cultural differences as stated above.

Conclusion And Implications:

In conclusion, high quality evidence regarding obstetrical outcomes and complications after vaginal delivery in women with a medical history of anorectal malformation is lacking. Up till now, recommendations on mode of delivery are based on low quality evidence derived from data that are poorly reported for many outcomes of interest or based on expert opinion. This paucity indicates the necessity of large studies investigating the obstetrical outcomes and complications in women with surgically corrected anorectal malformation. Furthermore, the development of core outcomes sets in this specific patient group should be developed.

Abbreviations

ARM
anorectal malformations
CS
cesarean section
OASI

Declarations

Ethics approval and consent to participate: Not applicable

Consent for publication: Not applicable

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Competing interest: The authors declare that they have no competing interest

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Authors' contributions: RG, CF, JJ, JD were involved in the design of this systematic. AW and JK performed the systemic review. AW and TA screened all articles and performed the data-extraction. RG, JJ and JD/CS were the third and fourth reviewer on the screening and data-extraction. Results were discussed among all authors. AW, RG, CF, and JD were major contributors in writing the manuscript. All authors read and approved the final manuscript.

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Figures

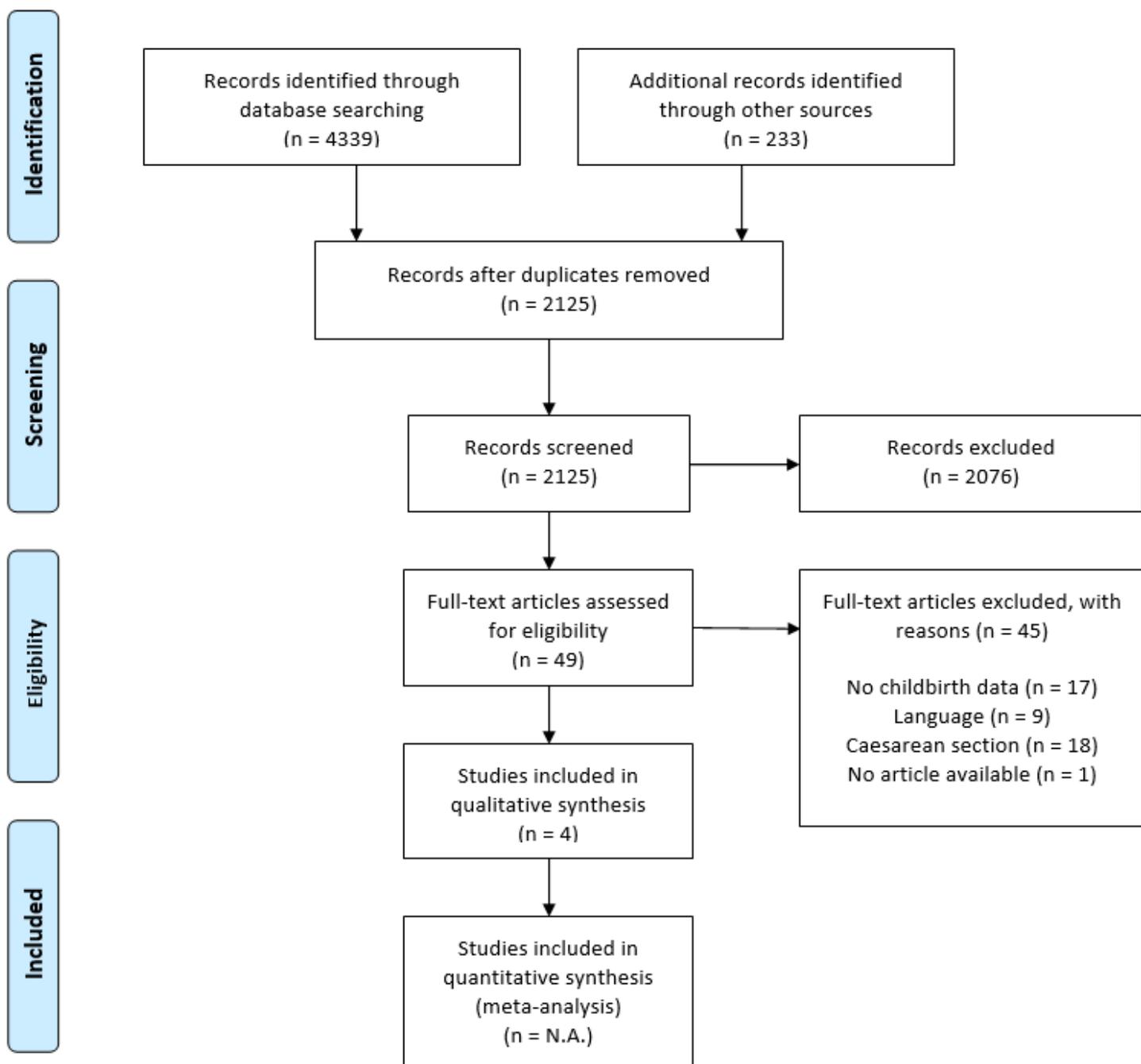


Figure 1

Prisma flowchart

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