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#### **Original Research Article**

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# Associated heart malformations in Iranian newborns with imperforate anus: A systematic review and meta-analysis

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#### **Abstract**

**Introduction**: Closed anus disease is a congenital disease in which the infant is not able to defecate normally. The aim of this study is that evaluated the prevalence of the heart malformations in Iranian newborns with imperforate anus . **Methods:** The methods used in this systematic review are developed based on the Checklist Guidelines (PRISMA). We included cross-sectional studies, case studies, and cohort studies, and excluded case studies, letters to editors, case reports, clinical trials, study protocols, systematic reviews, and reviews.

**Results:** A total of 251 articles were extracted through initial searches in various databases. A total of 779 patients were evaluated. All studies were retrospective studies. Based on the random effect model, the total Prevalence ofheart malformations in 779 patients was 26% (95% confidence interval and 23%, 29%).

**Conclusion:** Recognition of congenital heart disease before, during and after closed anal surgery helps a lot in the treatment process of these patients and nursing care.

**Keywords:** Imperforated anus, Associated anomalies, Frequency, heart malformations, malformations

#### Introduction

Congenital heart disease is a kind of heart disease that a baby is born with that (1). Closed anus disease is a congenital disease in which the infant is not able to defecate normally (2,3). The prevalence of congenital heart disease in the normal population is 5.8 per 1,000 live births ,but in patients with anal atresia about 25% have been (4).The anal atresia significant reported association with congenital heart disease and other abnormalities such as brain abnormalities, extremities, gastrointestinal, diaphragmatic hernia, face and ear anomalies,

pancreas, vision, hearing and so on (5). The high type is more associated with congenital heart disease and numerous other anomalies (6). Various heart diseases such as Trancus arteriosus, pulmonary artery atresia, dextrocardia and common types such as tetralogy of Fallot and VSD (ventricular septal defect) have been reported (7). Obtaining information about the presence of congenital heart disease in patients with closed anus is effective in the clinical treatment of these patients and the results of surgery, gave better results from surgery of

patients with closed anus(8). Anal abnormalities are one of the most common congenital malformations that are the result of defective development(9). The disease covers a wide range of abnormalities from simple closed anus to complex abnormalities of the pelvic and urogenital organs. One of the most common types of these abnormalities is anomaly, which is a type of abnormality in which the anus is not clearly open and there is often a fistula from the distal rectum to the perineum or urogenital system (10).

#### **Methods**

#### **Inclusion criteria (eligibility criteria)**

The methods used in this systematic review are developed based on the Checklist Guidelines (PRISMA). We included cross-sectional studies, case studies, and cohort studies, and excluded case studies, letters to editors, case reports, clinical trials, study protocols, systematic reviews, and reviews.

**Participants**: All studies on the Prevalence ofheart malformations in Iranian newborns with imperforate anus.

**Sampling methods and sample size:** All observational studies, regardless of their design, were included in the systematic review. The minimum sample size was 25 patients or more.

#### **Search strategy**

The searches were conducted by two independent researchers and the purpose of the search was to find published studies from 1/1/2000 to 5/30/2019. Studies published in MEDLINE were searched through PubMed, EMBASETM through Ovid, the Cochrane Library, and the English Trip database. Systematic review articles using MESH phrases and open phrases in accordance with print standards. After the MEDLINE strategy was finalized, the results were compared to search other databases, and PROSPERO was searched for recent or ongoing systematic reviews. The keywords used in the search strategy were:

Imperforated anus, Associated anomalies, Frequency, heart malformations, malformations

#### Select study and extract data

The two researchers independently analyzed the titles and abstracts of the articles according to the eligibility criteria. After eliminating additional studies, the full text of the studies was collected based on the eligibility criteria and information about the authors if necessary. General information (relevant author, province and year of publication), information about the study (sampling technique, diagnostic criteria, data collection method, research conditions, sample size and risk of bias) and output scale were collected.

#### **Quality evaluation**

The developed scale of Hoy et al. Was used to assess the quality of the method and the risk of bias in each observational study. This scale collected 10 items to evaluate the quality of studies according to their external validity (items 1 to 4 of the target population, sampling framework and minimum participation bias) and internal validity.

#### **Results**

#### **Study selection**

A total of 251 articles were extracted through initial searches in various databases. Out of 251essential studies identified by analyzing titles and abstracts,212 studies were omitted due to irrelevant titles. 25 articles were removed from 39 existing studies. Out of the remainingstudies, 6 studies met the study criteria. (figure 1).

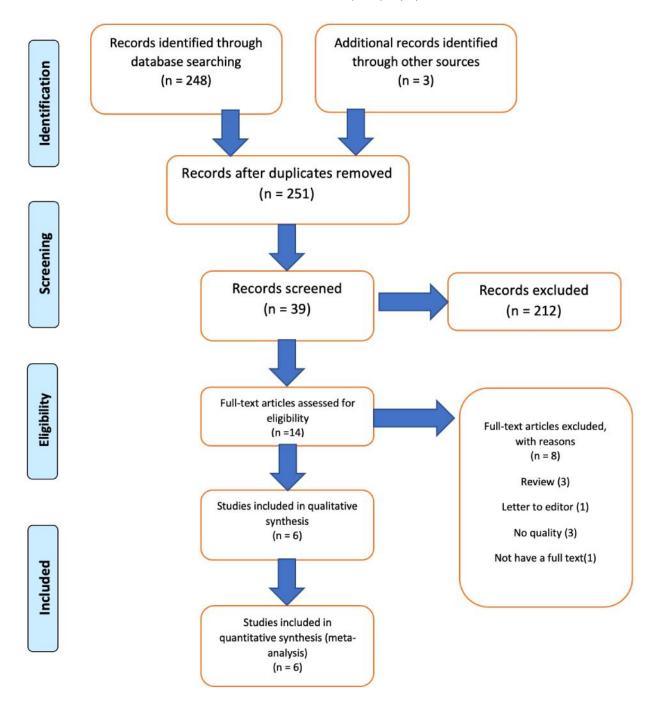


Figure 1:PRISMA flow diagram

#### **Research specifications**

A total of 779 patients were evaluated. All studies were retrospective studies. A total of 6 studies from 6 provinces that met the inclusion criteria were reviewed. Studies were from Tehran,

Gorgan, Mazandaran, Mashhad, Isfahan, Tehran and Tabriz. The risk of bias was low in most studies. The main method of data collection was medical records. The main study sites was hospital(Table 1).

Table 1.characteristics of included studies

Author	Year	Participant	Province	Mother's Age	Bias	Prevalence
Hashemi <sup>20</sup>	2012	97	Mashhad	NA	Low	0.30
Rashidi <sup>21</sup>	2006	90	Mazandaran	24±3	Moderate	0.54
Mirfazeli <sup>22</sup>	2014	40	Golestan	NA	Low	0.49
Davari <sup>23</sup>	2010	128	Isfahan	NA	Moderate	0.06
Delshad <sup>24</sup>	2010	245	Tehran	NA	Low	0.18
Hanife <sup>25</sup>	2016	179	Tabriz	NA	Low	0.72

#### Meta-analysis of the Prevalence of heart malformations in Iranian newborns with imperforate anus:

Based on the random effect model, the total Prevalence of heart malformations in 779 patients was 26% (95% confidence interval and 23%, 29%) (Figure 2).

#### **Subgroup analysis:**

#### **Meta-regression results:**

Results of meta-regression between male to female ratio and Prevalence of heart malformations in Iranian newborns with imperforate anus:

The meta-regression was evaluated according to the relationship between the Prevalence of heart malformations and male to female ratio. There was no significant linear trend in univariate metaregression to explain the change in the effect of age of participants. (Figure 3).

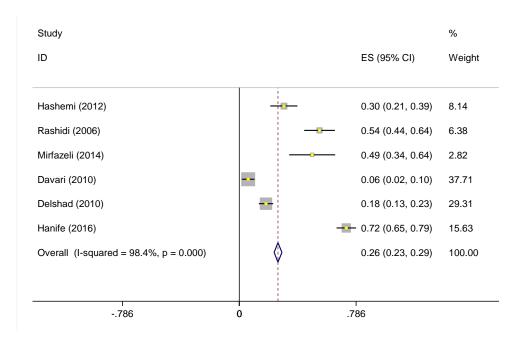


Figure 2.Meta-analysis of the Prevalence of heart malformations in Iranian newborns with imperforate anus

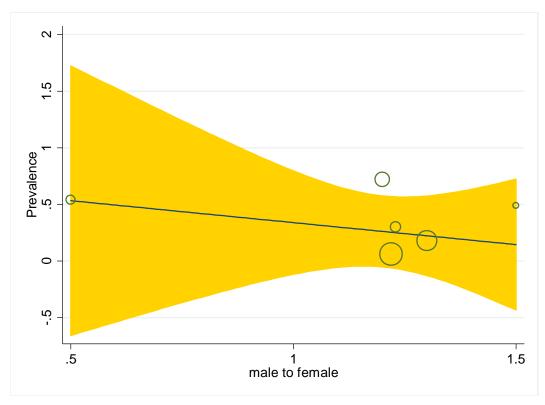


Figure 3 : Meta-regression between male to female ratio and Prevalence ofheart malformations in Iranian newborns with imperforate anus

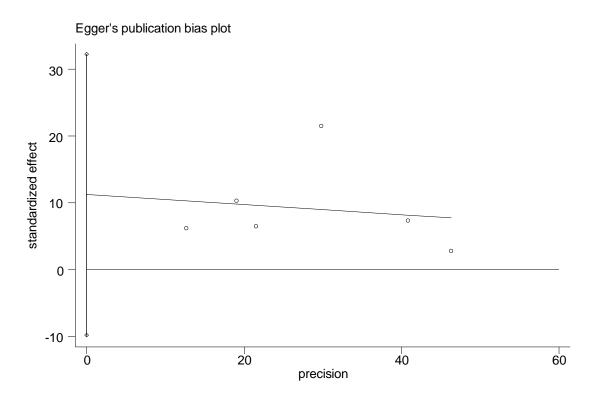


Figure 4.Egger's publication bias of the Prevalence of heart malformations in Iranian newborns with imperforate anus

#### **Conclusion**

Based on the random effect model, the total Prevalence ofheart malformations in 779 patients was 26% (95% confidence interval and 23%, 29%) .This fistula may be detected when meconium is found in the vaginal opening or below the vagina in the male urethra or perineum(11). The presence of meconium in the perineal area is not a reason for the anus to be open. The fistula may be indistinguishable at birth, but as the bowel movements increase, the meconium progresses through the fistula to the infant's urethra or perineum(12). By implementing a bowel control program, these children may achieve voluntary defecation at a socially acceptable level(13).Also, due to the high prevalence of congenital heart disease in people with closed anus compared to the general population, it seems that echocardiography is necessary in all patients, especially in terms of predicting the outcome of surgery and preventing possible complications(14). Environmental factors such as maternal diabetes play a role in its development.Rectal-urethral fistula, followed by perineal fistula, is the most common type of defect in male infants(15).In both sexes, an unperforated anus occurs in less than 5% of cases in the absence of a fistula and is often associated with Down syndrome(16).Babies with closed have congenital anus may also other abnormalities, including spinal disorders, anal atresia, congenital heart disease, esophageal fistula, esophageal atresia, kidney problems, radial anomalies, and limb defects(17). The prevalence of congenital heart disease in the normal population is 5 to 8 people per 1333 live births(18).But in patients with closed anus, it has been reported up to about 25%. Knowing the existence of congenital heart disease in patients with closed anus is effective in the clinical treatment of these patients and the results of surgery(19). If the rate of this disease in patients with closed anus is significant, heart counseling can be performed before surgery. In the treatment process, performed the necessary intervention to obtain better results from the surgery of patients

with closed anus.Recognition of congenital heart disease before, during and after closed anal surgery helps a lot in the treatment process of these patients and nursing care.

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