IJCRT.ORG ISSN : 2320-2882

INTERNATIONAL JOURNAL OF CREATIVE RESEARCH THOUGHTS (IJCRT)

An International Open Access, Peer-reviewed, Refereed Journal

Acceptance of Premarital Counseling, Prenatal Diagnosis and Abortion of Affected Fetus by Parents of BetaThalassemia Children; A Systematic Literature Review.

Prof. Dr. N.H.Simon, Director at school of Health Sciences, Department of Public Health, Noida International University.Gautam Buddh Nagar, Uttar Pradesh, India.

Masuma Kazimi, MD, MPH student, Department of Public Health, Noida International University, Greater Noida, Uttar Pradesh, India.

Abstract

Introduction: Thalassemia, a hereditary autosomal recessive disorder, imposes a major public health burden in an area known as thalassemia belt. Thalassemia belt extends from Mediterranean region to middle east and south east Asia. To avert the fiscal and social burden on health system, countries have devised and adopted preventive strategies. Although, WHO developed and disseminated a preventive strategy for prevention of hemoglobinopathies, including thalassemia, countries must adjust it according to the prevailing sociocultural determinants within countries.

Objectives: This study aims to investigate acceptance of the preventive measures for thalassemia i.e. premarital counseling, prenatal diagnosis and abortion in existing body of literature. Furthermore, it seeks barriers towards successful implementation of these preventive approaches.

Methodology: Online search engines (Google scholar and PubMed) were used for exploring related studies that meets the inclusion criteria. Studies conducted from 2000 onwards were illegible for the study.

Result: With strictly following PRISMA methodology, 11 studies were included for the full-text reading from the total of 4097 articles. The results of studies varied widely; for instance, the acceptance of premarital counseling ranged from about 59% to 84% by different studies that had been investigated fully. Similarly, Acceptance of prenatal diagnosis was reported from 100% to as low as 74% by studies conducted in India. This study found out that acceptance of premarital counseling was higher in Muslim majority countries compared to PND and abortion whereas the opposite is true in India. Furthermore, preventive strategies and social mobilization can significantly transform the acceptability of prohibited preventive measures such as PND (El- Beshlawy et al, Egypt).

Conclusion: With the wide discrepancies found in the results of studies that have been conducted, further investigations are crucial to reflect the true acceptance of each of preventive approach within India. Although, the results of the study highlights that Muslim communities are in favor of premarital counseling whereas prenatal diagnosis and abortion of affected fetus is welcomed in multiethnic countries like India. This will contribute in successful development of strategies that can halt the raising trend of thalassemia in India effectively.

Key words: Thalassemia prevention, premarital counselling, prenatal diagnosis, Thalassemia parents.

1.Introduction

Thalassemia, a hereditary hematological disease, is highly preventable. Prevention of new births of the thalassemia can be ensured through screening, genetic counseling in combination with prenatal diagnosis (PND)

and abortion of an affected fetus [1]. This approach is cost-effective and is proving remarkably successful in reducing the frequency of thalassemia in many countries [2-3]. While counties such as Cyprus and Iran have achieved considerable reduction of thalassemia cases through implementation of premarital genetic counselling and prenatal genetic diagnosis, it is still a major problem in India. Existing literature suggests that the best approach for combating the disease varies from one culture to another because of the disparate acceptability of preventive approach.This is because marriage is a complex social phenomenon, and the selection of marriage partners are based on a strong personal preference, family or traditional reasons. It is stated that unlike Iran and Cyprus, Indian communities are less receptive towards premarital genetic counselling, rendering prenatal diagnosis in spotlight for preventive measures [4].Hence in the endeavor to halt and reverse the trend of thalassemia, the acceptability of these approaches must be evaluated. It is important to find feasible approaches for prevention since despite noticeable efforts for the provision of screening and genetic counselling services, India is still struggling with the burden of thalassemia.

2.Methodology

Online search engines [google scholar and PubMed] were used for screening the existing literature. All types of study designs were included in condition to be conducted from 2000 onwards. While the priority was given to the research conducted in India, countries within Thalassemia belt (in middle and South East Asia) were included. Keywords used for search engines were: prenatal genetic diagnosis, premarital genetic counselling, and attitude towards beta thalassemia. The articles were selected through using PRISMA model of systematic review. The PRISMA flowchart depicting the systematic search used in this article is illustrated in Diagram 1.1

Table 2.1 inclusion and exclusion criteria for the study.

No	Criteria	Inclusion criteria	Exclusion criteria
1	Date	2000– 2020	Studies conducted before 2000
2	Location	Countries located in the south and middle east Asia.	All countries other than those mentioned in included countries
3	Gender	Female and male	
4	Language	English	Languages other than English
5	Study design	All types of study designs other than review articles	Review study
6	Study area	Entailing studies about attitudes of high risk beta thalassemia towards prenatal and premarital genetic counselling for beta thalassemia prevention.	Studies concerning about other preventive measures apart from genetic counselling and related to other subtypes of thalassemia such as alpha thalassemia and sickle cell and thalassemia coexistence.

Research question designed for this research

- What is the acceptability of prenatal diagnosis among people?
- What are barriers against adoption of prenatal diagnosis?
- How likely parents of a thalassemia child encourage their extended family to practice premarital counseling.
- Whether parents accept termination of an affected fetus following prenatal diagnosis.

PRISMA SYSTEMATIC RIVIEW FLOWCHART

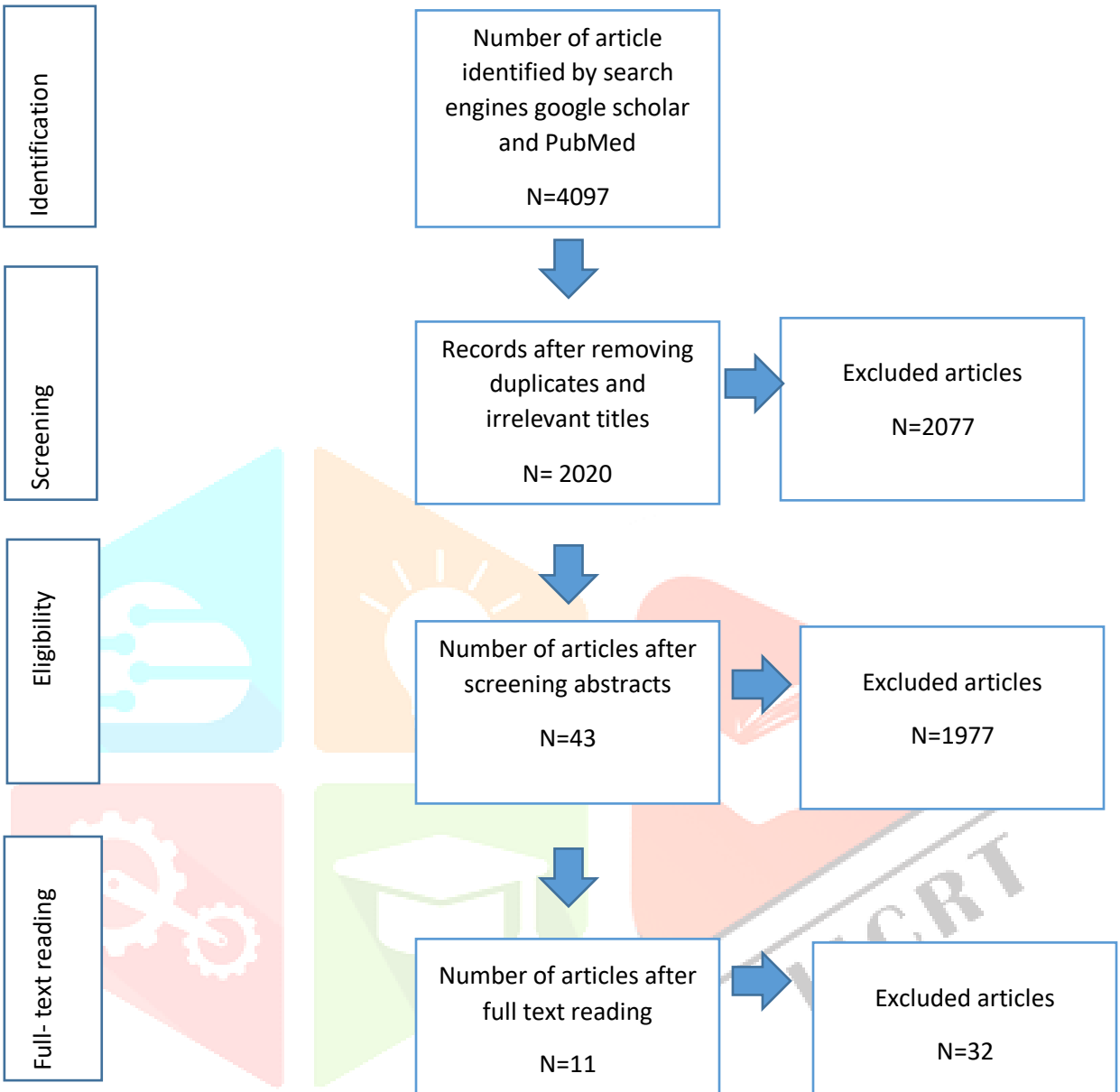


Diagram 1.1 PISMA Figurative Illustration

2.1 Data extraction table

No	Author	Title	Study Design	Sample Size	Study population	Results
1	Dr. Sarita Agarwal et al. (2002), India	Prenatal diagnosis in beta thalassemia: an Indian experience	experimental	53 couples	Couples with one affected child	29 % (14 pregnancies) thalassemia affected fetuses diagnosed via PND were all aborted; hence, the acceptance of PNC was 100 % following PND. All patients were amenable to attend PND counseling to avoid another thalassemia child.
2	Parag M. Tamhankar et al. (2009), India	Prevention of thalassemia by premarital screening and prenatal diagnosis in India	experimental	1348 Anemic OPD patients 939 college students 400 extended families	OPD patients College students Extended families	99 percent of identified high risk couples proceeded with the marriage. However 66.5% of these couples admitted for PND,whereby, 33 births of thalassemia children were avoided. After councelling From the recognised high risk couples 74.3, 37.5 and 100 percent of extended family members, OPD and college students admitted for PND , respectively (64.7% overall admission rate).
3	Asha Baxi et al. (2012), India	Carrier screening for beta thalassemia in pregnant Indian women	experimental	1320	Pregnant women in ante natal care	76 percent of pregnant women accepted screening for beta thalassemia, 99 percent of which were willing for PND if required.
4	Swati Chawla et al (2017), India	Attitudes and beliefs among high- and low-risk population groups towards β-thalassemia prevention	cross-sectional descriptive study	926	Arura Rural (AR) population from both male and female from house to house visits from Rohtak	73.3 % were amenable towards screening for beta thalassemia. Moreover, while 59.4 % conceded that they would do premarital genetic testing, 76.3 % accepted PND if required. Acceptance of abortion following PND was 67.3%. Higher proportions of AU (54.7%) are not willing for premarital screening than those of AR and CC. Furthermore AR preference for abortion is the highest in (96.4%) followed by AU (72.9%) and CC (62.9%). A great

					District, Haryana state in north India, and Arora Urban (AU) and cosmopolitan commoner (CC) from New Delhi Kalanur village of	numbers of CC have willingness to share knowledge about β-thalassemia with others, whereas half of the AR and AU groups are not willing to share the same.
5	Jahnavi Hatti (2015), India	Genetic counseling in Anemia and Thalassemia	Cross Sectional Study	71	Parents of beta thalassemia patients	Consanguinity was detected in 42 (59%) of cases whereas 29 (40.84%) was non consanguineous marriages culminating in Thalassemia children 30 percent of families did not opted for prenatal counseling because they considered the given information inadequate however, 11 percent did so for the fear of stigma. 24 and 35 percent of those who refused prenatal diagnosis had misconception of cost and financial hardships respectively.
6	Ahmed I. Gilani et al. (2007), Pakistan	Attitudes towards Genetic Diagnosis in Pakistan: A Survey of Medical and Legal Communities and Thalassemic Children parents	Cross sectional study	902	570 doctors, 49 lawyers, 178 medical students, 89from parents of thalassemic children and 16 members of parliament (MPs)	11% did not considered genetic screening acceptable of which 38.3%did so due to prioritization of other morbid diseases, 26% and 34% did so because of stigmatization and stimulation of a surge in abortion rate respectively. 77% were in favor of premarital genetic screening and 94.4% of parents were in favor of prenatal screening, compared with 77.4, 51 and 70.8% of doctors, lawyers and medical students, respectively, In case of a scenario of a child expected to have a life of disability due to a genetic disease only 1 MP (6%) was in support of any sort of prenatal screening.

7	Fouzia Ishaq et al. (2009), Pakistan	Awareness Among Parents of β-Thalassemia Major Patients, Regarding Prenatal Diagnosis and Premarital Screening	Descriptive cross sectional study	115	Parents of beta thalassemia patients Comprising of 74 men and 41 women	97 (84.3%) and 88 (76.5%) parents were aware of Premarital screening and prenatal diagnosis respectively. From Ninety-nine parents (86.1%) who were knowledgeable about the termination of pregnancy on positive prenatal test only 69 considered it acceptable religiously.
8	El-Beshlawy et al (2012), Pakistan	Prenatal diagnosis for thalassaemia in Egypt: what changed parents' attitude?	Experimental study	71	pregnant mothers at risk for β-thalassaemia	Twenty-four women (33.8%) were found to have affected fetuses; 100% of these women opted to terminate the pregnancy. The change in attitude towards termination of pregnancy was related to in-depth counseling of the religious believes aspects towards prenatal diagnosis and annulment of pregnancy
9	Ching Fang Ngim et al (2013), Malaysia	Attitudes towards prenatal diagnosis and abortion in a multi-ethnic country: a survey among parents of children with thalassaemia major in Malaysia	Cross sectional study	116	parents of children suffering thalassemia major	The majority (83/71.6 %) had willingness for prenatal diagnosis, but only 33 (28.4 %) agreed to both prenatal diagnosis followed by termination of affected fetus 73.4% of Muslim participants were against termination compared to 25% of Christians and 13.3% of Buddhists and 100% Hindus
10	Mehran Karimi et al	Attitude toward prenatal diagnosis for β-thalassemia	Cross-sectional descriptive study	764	Parents of thalassemia children (66.7%) and	While 711 (93%) were in favor of prenatal diagnosis, 53 (6.9%) were not. Specifically, 663 subjects (86.8%) were in favor of early termination of pregnancy in case of an affected fetus, while (13.2%) were not

	(2006-2007), southern Iran	major and medical abortion in southern Iran			thalassemia patients (33.2%).	
11	Ayman Alsulaiman et al (2006), Saudi Arabia	Attitude determination to prenatal and preimplantation diagnosis in Saudi parents at genetic risk	Cross sectional study	30	Families with a child with morbid thalassemia gene	Eight of the 30 couples (27%) would only accept PGD; four (13%) only PND; three (10%) either technology; the remaining half (50%) would accept neither test, or were undecided. Twenty three parents declared disagreement with abortion following PND and stated religious reasons as driving factor.

Abbreviations: prenatal diagnosis (PND), prenatal genetic counselling (PNC), outpatient department (OPD), urban and rural Arora (UA & RA respectively), cosmopolitan commoners (CC).

2.2 Quality assessment of studies using AXIS (Appraisal Tool for Cross-sectional Studies) guidelines

From the 11 studies that were included in the article, 7 were cross- sectional study which, for quality assessment (QA) purpose, the AXIS guideline was used. AXIS tool examines survey articles in terms of their introduction, methodology, result, discussion and conclusion and other (funding and conflict of interest, and consent). The aims and hypothesis are scored one each in introduction (total 2 scores), similarly, appropriateness of study design, justification of sample size, generalizability of sample frame, representativeness of sample, definition of target population, validity, reliability, non-responders measures, explanation of statistical methods and statistical significance individually merit one score in methodology (total score of 10). Result carries 5 marks which is determined based on clearly explanation of results, concerns about non-responders, measurements of non-responders, consistency of results and the compliancy with methodology. Furthermore, whether the study's discussion and conclusion was in accordance with results and whether it mentioned the limitations of the study a total score of 2 was assigned for each respective part. Finally, one score is given if either the funding or conflict of interest was mentioned, besides, one extra one mark was allotted for the consent form for respondents in the study. Eventually, the highest score a study can get is 21 marks which 9 is the yardstick for a study to be considered qualified in this article. Once studies were assessed using Axis tool, one scored below the quality standard while the others were found to be quality assured. The results are depicted in the table 2.3

Quality assessment of cross-sectional studies using AXIS guideline

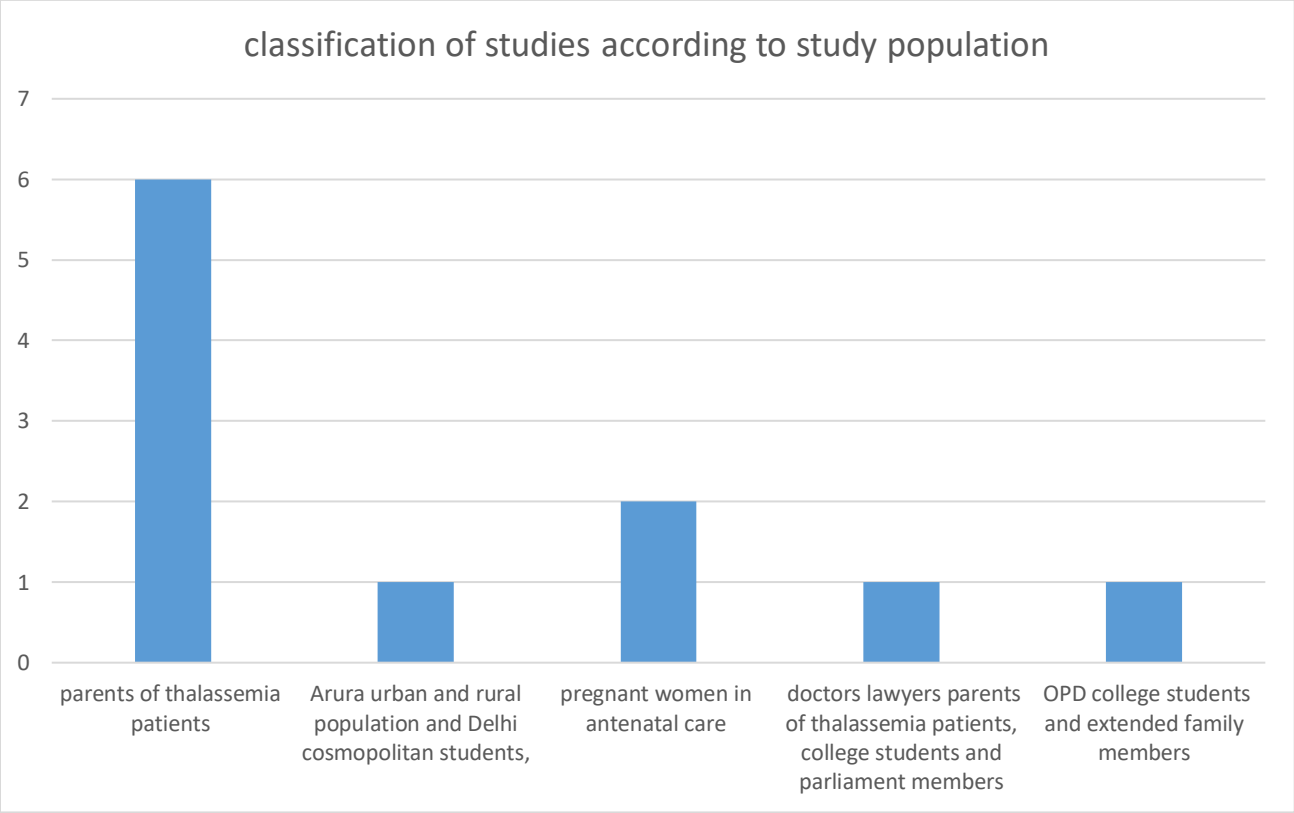
Name of the author /year of publication	Introduction (2scores)	Methods (10 scores)	Results (5 scores)	Discussion (2scores)	Other (funding source, conflicts of interest, ethical approval) (2score)	Total (21scores)
Swati Chawla et al 2017	2	8	3	2	2	17
Jahnavi Hatti et al 2015	1	4	1	0	0	6
Ahmed I. Gilani et al. (2007)	1	9	6	3	0	16
Fouzia Ishaq et al. 2009	1	6	2	2	1	12
Ching Fang Ngim et al 2013	1	4	4	2	1	12
Mehran Karimi et al 2006-2007	1	4	3	1	2	11
Ayman Alsulaiman et al 2006	1	3	4	1	1	10

3.Results:

4097 studies were retrieved from the search result, 4054 got excluded because the study tittles and abstracts did not address the research questions, 43 articles were assessed for eligibility from which 32 studies were excluded because they had inappropriate study population, did not include neither premarital nor prenatal genetic counselling. As a result, a total of 11 studies were recruited in this study

Five Studies were conductedin India. Dr. Sarita Agarwal et al (2002),Jahnavi Hatti(2015),Mehran Karimi et al (2006-2007),Ayman Alsulaiman et al (2006), Fouzia Ishaq et al (2009),Ching Fang Ngim (2013) conducted their studies among parents of thalassemia patients

Population study for Swati Chawla et al (2017) was Arura urban and rural population and Delhi cosmopolitan students, whereas Parag M. Tamhankar (2009) did his study among OPD patients, college students and extended family members. Pregnant women in antenatal care made up study population in studies conducted by Asha Baxi et al (2012) andEl-Beshlawy et al (2012). Ahmed I. Gilani et al (2007) included doctors, lawyers,and parents of thalassemia patients, college students and parliament members in his study. Study population and sample sizeof each study is shown in the bar graphs.



1.1 study population of included articles

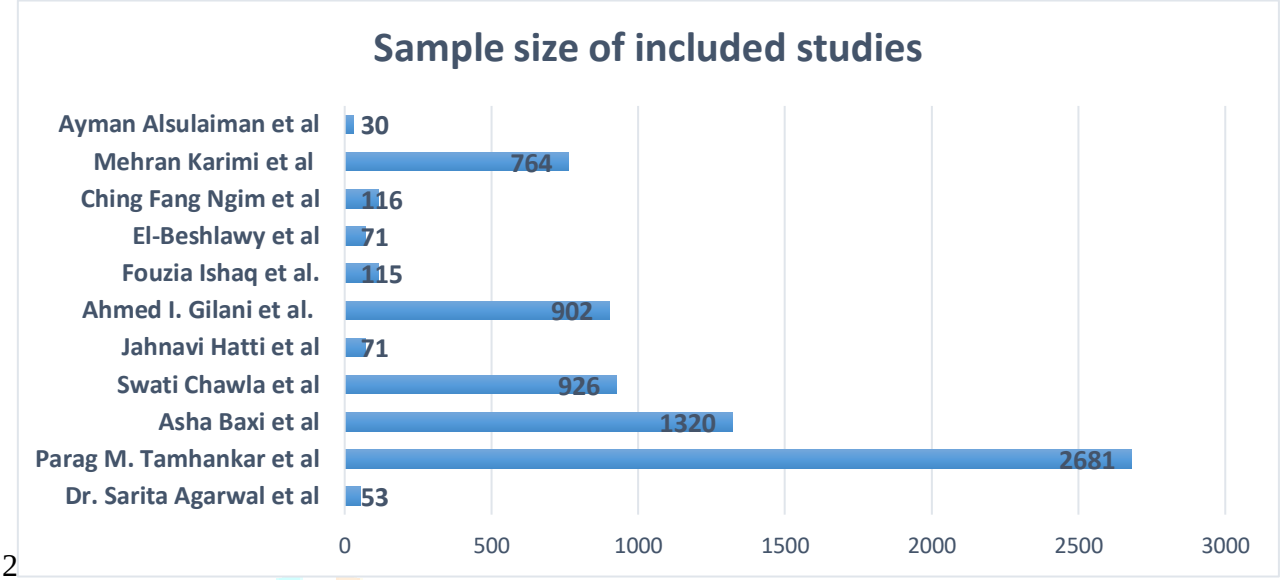


Table 1.2

4. Discussion

4.1 Perception and practice of premarital screening and counselling

From all studies entailed in this article two (Swati Chawla et al from India and Ahmed I. Gilani from Pakistan) reported willingness towards premarital genetic counselling for prevention of beta thalassemia. Out of 926 respondents in Swati Chawla's cross sectional study, 59.4% expressed adoption of premarital genetic counselling. Supporters of this initiative was reported higher (77%) by another cross sectional study by Ahmed I. Gilani in Pakistan. Fouzia et al (2009) reported awareness of premarital genetic counselling 84.3% among parents with a thalassemic child in Lahore, Pakistan.

4.2 What is the acceptability of PND among parents of thalassemia patients?

In all five studies that were conducted in India acceptance of PNDwas reported in four of them (100 % by Sarita Agarwal et al 64.7% by Parag M Tamhakar, 99% by Asha Baxi et al and 76.3% by Swati Chawla et al). Acceptance of PND was 71.6 percent in a study in Malaysia carried out by Ching Fang Ngim et al in 2003. 28.1% of Muslim participants were against termination of pregnancy compared to 50 % of Christians and 11.8% of Buddhists and 100% Hindu in that study. In a cross sectional study conducted by Ahmed I. Gilani et al in Pakistan Islamabad on 902 respondents of specified groups (doctors, parents, lawyers, members of parliament and medical students) found that 89% of respondents were receptive of the PND. In Iran, another Islamic majority country, Mehran Karimi reported acceptance of PND 93% through a cross-sectional study performed in southern Iran. In Saudi Arabia, Ayman Alsulaiman published the result of his study as 13 % were amenable toward prenatal diagnosis whereas 27 % were supportive of pre-implementation genetic counselling for prevention of beta thalassemia.

4.3 What is the acceptance of abortion following prenatal diagnosis for beta thalassemia major?

Both of studies conducted in India by Sarita Agarwal et al (2002) and Parag M Tamhankar et al (2009) showed a 100 percent acceptance of abortion among those who had undergone PND and the fetus was diagnoses to have beta thalassemia major. However, another study by Swati Chawla et al (2017) yielded that only 67.3 % of respondents were agreeable about abortion of an affected fetus. The former two studies are among parents with one affected child or were previously screened for beta thalassemia trait, the later on, the other hand, was conducted using door to door survey method. Fouzia et al(2009) under her cross sectional study in Pakistan reported 69% of population accepting abortion. While Mehran Karimi et al claimed that 86.8% of his subjects accepted early termination of affected pregnancy , Study of Ayman Alsulaiman in Saudi Arabia, another Muslim majority country, contended that 76.7% of 30 couples who had one affected child refused to terminate an affected

pregnancy. El-Beshlawy et al (2012) asserted that as a result of in depth counselling on religious aspect of abortion 100 percent of pregnancies with affected fetus adopted abortion as a means to evade a beta thalassemic child. He mentioned that a noticeable improvement had occurred in the attitude of parents towards termination of thalassemia major pregnancy since it was reported 36% acceptance of abortion by previous studies.

4.4 Socio-economic, religion and education influences

Parag M. Tamhankar (2009) in his study suggested that all college students who had screened for thalassemia admitted for prenatal diagnosis after marriage compared to extended family members and OPD patients (74.3% and 37.5% respectively). Moreover, Swati Chawla et al (2009) carried out a study among rural and urban Arura population and cosmopolitan commoners of New Delhi. Her findings were as follows: rural Arura with 81% acceptance of premarital screening was the highest in this term. Similarly, abortion was widely accepted among rural Arura (96.4) followed by urban Arura (72.9%) and cosmopolitan commoners (62.9%). In the study conducted by Ahmed I. Gilani et al. (2007) 77% were in favor of premarital genetic screening and 94.4% of parents were in favor of prenatal screening, compared with 77.4, 51 and 70.8% of doctors, lawyers and medical students, respectively, In case of a scenario of a child expected to have a life of disability due to a genetic disease Only 1 member of parliment (6%) was in support of any sort of prenatal screening..

A cross sectional study by Ching Fang Ngim et al (2013) in Malaysia yielded the results that reflect the religious constructs on the matter of prenatal diagnosis of thalassemia. He and his team reported that 28.1% of Muslim participants were against termination compared to 50 % of Christians, 11.8% of Buddhists and 100% Hindus.

5. Conclusion

There is less literature regarding acceptance of premarital counselling by parents of thalassemia children in India. In the past twenty year only one study that was conducted among residents of Arora by Swati Chawla, acceptance was 59 %. However it does not reflect the acceptability among parents of thalassemia children. Overall, the acceptance of premarital counseling is higher than PND and abortion while the opposite trend can be seen in India.

Acceptance of PND is reported with remarkable variations in the studies in India ranging from 64.7 to 100%. Similarly, it varies among Muslim majority countries (93% in Iran and 71.6% in Malaysia), indicating that other social factors determines the acceptability of PND in communities.

In order to effectively halt the rising trend of thalassemia, PND must be adopted only when the acceptance of abortion of a severely diseased fetus is acceptable. The acceptance of abortion among parents of beta thalassemia children is reported high in studies by Sarita Agarwal et al (2002) and Parag M Tamhankar et al (2009), while in studies in other countries the acceptance was significantly lower. This shows the need for re-assessment of the acceptance of abortion among Indians. The relationship between socioeconomic factors and acceptability of premarital counselling and prenatal diagnosis has not been investigated enough since in the study done by Swati Chawla et al. rural area showed a higher acceptance compared to urban area and no other study has been repeated to confirm its validity and the underlying cause.

References

1. Maheshwari M, Arora S, Kabra M, Menon S: Carrier screening and prenatal diagnosis of ß- thalassemia. Ind J Pediatr 1999;36:119–125.
2. Thakur C, Vaz F, Banerjee M, Kapadia C, Natrajan PG, Yagnik H, Gangal S: Prenatal diagnosis of ß-thalassemia and other haemoglobinopathies in India. Prenatal Diagn 2000;20:194–201.
3. Ahmed S, Saleem M, Sultana N, Raashid Y, Waqar A, Anwar M, Petrou M: Prenatal diagnosis of B-thalassemia in Pakistan: Experience in a Muslim country. Prenatal Diagn 2000;20: 378–383.

4. Verma IC, Saxena R, Kohli S. Past, present & future scenario of thalassaemic care & control in India. Indian J Med Res. 2011;134(4):507-521.
5. S.Agarwal et al. prenatal diagnosis in beta thalassemia, an Indian experience. Fetal Diagnosis and Therapy. 2003; 18:328–332 DOI: 10.1159/00007197.
6. Parag M Tamhankar et al. prevention of homozygous beta thalassemia by premarital counseling and prenatal diagnosis in India. Wiley InterScience; 2008.
7. Asha Baxi et al. Carrier Screening for beta Thalassemia in Pregnant Indian Women: Experience at a Single Center in Madhya Pradesh. Indian Society of Hematology & Transfusion Medicine; 2012.
8. Jahanvi Hatti et al. Genetic counseling in Anemia and thalassemia. Acta Medica. Issue 1; 2015.
9. Swati Chawla et al. Attitudes and beliefs among high- and low-risk population groups towards β-thalassemia prevention: a cross-sectional descriptive study from India; Springer-Verlag Berlin Heidelberg 2017.
10. Fouzia et al. Awareness Among Parents of β-Thalassemia Major Patients, Regarding Prenatal Diagnosis and Premarital Screening; Journal of the College of Physicians and Surgeons Pakistan 2012, Vol. 22 (4): 218-221
11. Ahmad I. Gilani et al. Attitudes towards Genetic Diagnosis in Pakistan: A Survey of Medical and Legal Communities and Parents of Thalassemic Children. Community Genet 2007;10:140–146
12. Ayman Alsulaiman and J. Hewison. Attitudes to prenatal and preimplantation diagnosis in Saudi parents at genetic risk; Wiley InterScience. 2006.
13. Ching Fang Ngim. Attitudes towards prenatal diagnosis and abortion in a multi-ethnic country: a survey among parents of children with thalassaemia major in Malaysia, Springer-Verlag Berlin Heidelberg 2013.
14. A. El-Beshlawy et al. Prenatal diagnosis for thalassaemia in Egypt: what changed parents' attitude. John Wiley & Sons, Ltd; 2012.
15. Mehran Karimi et al. Attitude toward prenatal diagnosis for b-thalassemia major and medical abortion in southern iran. Hemoglobin, 34(1):49–54, (2010).