Knowledge, attitude, and practice of reproductive behavior in Iranian minor thalassemia couples

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ABSTRACT

الأهداف: الاهتمامات بالسلوك التناسلي للزوجين ذوي الخطر من الإصابة بالثلاسيميا الكبرى (TM).

الطريقة: أجري فحص وصفي على شريحة عرضية خلال عام 2006م – في جامعة مازندار الطبية – إيران. تم تسجيل معدلات الولادة خلال الفترة مابين عام 1997م وحتى عام 2005م، وعدد المرضى الجدد المسجلين عند مرحلة الخطر من الإصابة لدى العائلات. كانت الطريقة المستخدمة لجمع البيانات هي الاستبيانات المحتوية على الإحصائيات السكانية الجغرافية من الأزواج، المعرفة (20 سؤال) والوضع الجسماني (20 إفادة)، الممارسة عن طريق دراسة ملف العائلة في المركز الصحي. تم تعبئة الاستبيانات بواسطة الزوجين بشكل منفصل. تمت مقارنة الأعداد المتوقعة المضادة الفعلية من المرضى الذين تمت ولادتهم في نفس الفترة. كما تم تحليل البيانات بواسطة طريقة التحليل الإحصائي (SPSS13.00)، نسبة الخطأ ($p<0.05$) كنسبة ملحوظة.

النتائج: تمت دراسة 100 زوج من 240 زوج عند مرحلة الخطر. لدى 82% منهم معرفة جيدة عن مرض الثلاسيميا (TM)، والوضع الجسماني الايجابي كان 68.5%. كانت علاقات المعرفة مع الوضع الجسماني ملحوظة ($p<0.001$). لم يكن لدى 50% من الأزواج ممارسة غير مرغوبة. من دون PND، 4 مرضى يعانون من الثلاسيميا (TM) كانوا قد ولدوا، تم التبليغ عن 98 عارضة من الممارسة الغير مرغوبة. خلال هذه الفترة، تم استخدام مانع الحمل من قبل 12% من الأزواج بطريقة غير آمنة. كان عدد المرضى المشتبه إصابتهم بالثلاسيميا (TM) وليس لديهم برنامج وقاية 25 مريضاً. لذلك تم منع ولادة اثنان بالثلاسيميا (TM) (تقليص بنسبة 75%).

خاتمة: تم تحقيق إنجاز عظيم خلال الأعوام التسعة الماضية للتقليص من الإصابة بالثلاسيميا (TM) في هذه المنطقة.

Objectives: To investigate the knowledge, attitude, and practice of reproductive behavior in Iranian minor thalassemia couples in Ghaemshahr City, Mazandaran, Iran.

Methods: This is a cross-sectional descriptive survey conducted in 2006. Birth rates from 1997-2005 and the number of newly registered patients from at risk couples was recorded. Tools for data collection were a valid questionnaire containing epidemiologic characteristics of couples, knowledge (20 questions), attitude (20 statements), and practice (by studying the family file in health centers). Questionnaires were completed by husband and wife separately. Actual versus expected numbers of patients born in that period were compared. The data were analyzed using the Statistical Package for Social Science version 13.00, and $p<0.05$ was interpreted as significant.

Results: Of the 240 at risk couples, 100 were studied. Of them, 82% had good knowledge of thalassemia, and 68.5% had a positive attitude toward thalassemia prevention program. Correlations of knowledge with attitude were significant ($p<0.001$), and 50% of the couples had unfavorable practice including unplanned pregnancy, fetal abortion without prenatal diagnosis (PND), delivery without PND, and having a child affected by thalassemia major (TM). Without PND, 4 TM patients were born. Ninety-eight episodes of unfavorable practice were reported. Meanwhile, the contraceptive method used by 12% of couples was unsafe. Suspected TM patients with no prevention program were 25; thus, the birth of 2 TM was prevented (92% reduction).

Conclusion: We achieved great success during the last 9 years in the region, and TM prevention program improved knowledge, attitude, and practice in high-risk couples and carrier families.

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National efforts for prevention of thalassemia major (TM) began at least 2 decades ago.[1,2] In Iran, Mazandaran province was always a pioneer in this regard.[3,4] One of the most important strategies for prevention is pre-marital screening and genetic counseling of carrier couples, which has been practiced since 1991.[1] Screening in the area as a national program, started in the autumn of 1997. Carrier couples are counseled on TM and the chance of having affected children. Approximately 11% of Mazandaran residents are beta thalassemia carriers (beta-thalassemia minor), and in 2% of married couples (1% of couples who had married before are both carriers). Also, approximately 50% of carriers decided to marry, so there are many at risk families living in the region.[3,4] Couples who decide to marry need to participate in special educational sessions that include information on their situation, risk of having affected children, TM disease, and its possible management. Family planning, prenatal diagnosis, and legal abortion of affected fetuses is also discussed. Then they are included in a supervision program regarding family planning and their method of contraception.[1-4] Records of the couples were kept in the health center, and periodic visits are performed by health workers. Prenatal diagnosis (PND) and abortion of the affected fetuses is currently available, and the legal/economic aspects are supported by insurance companies.[2,5-7] Nevertheless, years after the initiation of the program, there are concerns on couples present knowledge and attitude toward the disease, and also their happiness regarding decisions made, and their practice regarding contraception, PND, and abortion of affected fetuses. The aim of this study was to evaluate the knowledge, attitude, and reproductive practice behavior of minor thalassemia couples.

Methods. This is a cross-sectional descriptive study conducted in 2006 in randomly selected health network of Ghaemshah province. The list and addresses of couples with the following characteristics (at risk families) were extracted from the health center records: 1) carrier state of both partners and 2) living together for at least one year. Also, birth rates from 1991-2005 as well as the number of newly registered TM patients from supervised at risk families were recorded from the health center statistics. The study questionnaire for assessing the knowledge and attitude of the couples was designed by a research team including an expert physician involved in counseling and education of at risk couples for many years and 2 pediatricians with appropriate history of practice in the thalassemia ward (content validity). The reliability of the questionnaire was controlled by the test-retest method (r: 0.8). The questionnaire was given to the family and we asked the husband and the wife in each family to complete the form separately and confidentially. In the covering letter, the word confidential was stated. The epidemiologic characteristics of the couples were assessed by 10 questions. The knowledge part by 9 multiple choice plus 11 true or false questions, with a total score of 20. Patients with scores of 75% were interpreted as "good knowledge" and scores between 75-50% were labeled as "acceptable knowledge." Also, the attitude was assessed using 20 statements, which were scaled using 4 options Likert scoring system: 1 - absolute agreement, 2 - relative agreement, 3 - relative disagreement, and 4 - absolute disagreement. Obtaining 1-4 quartiles of scores was interpreted as absolute and relative positive and absolute and relative negative attitudes. Practice regarding prevention of TM in offspring was assessed via the family records in the health center. Any of the following situations alone or in combination were considered as unfavorable practice, including unplanned pregnancy, fetal abortion without PND, delivering a child without PND, and having a child affected by TM. The contraceptive method used by the couples during the period was reported according to the file name as safe (tubal ligation, vasectomy, menopause, intrauterine device [IUD], pill) or unsafe (withdrawal/abstinence methods). The socioeconomic status of the families was assessed by 8 variables covering: academic educational level of parents, financial carrier categories of both spouses, place of residence, and possession of residential place. Zero to grade 3 were given to each category regarding their role in family income and social class, and then the socioeconomic status of each family was defined. Employed women and those with university degrees received higher points than similar categories for men. Actual affected births were compared to expected numbers according to birth rates in the corresponding years. The studies were fully explained to the patients and informed consent was obtained. All the study protocols were approved by the medical ethics committee of Mazandaran Medical University.

Descriptive statistics were used to obtain the data, and the correlation of some variables was studied using Spearman Chi square test. Odds ratio and 95% confidence interval (CI) were calculated. A p value of <0.05 was interpreted as significant.

Results. One hundred registered families were asked to complete the questionnaires. Of them, 52% were residents of a rural area. The age of the women was 26.5±7.5 years (range 14-51) and of men was 30.4±8.6 years (range 20-75). The couples were living together for 4.3±3 years (range: 1-9). The first genetic counseling was performed 2.8±2 years (range: 1-9)

before the marriage. Twenty-nine participants (14.5%) could not remember participation in a counseling session before marriage. We assumed that 25% of the couples completed the questionnaire separately, all the 200 filled-up questionnaires were collected. Of these, 75 couples were not genetically related, 17 were first cousins and 12 were related, but not first cousins. Regarding formal educational level: 105 (52.2%) did not finish high school, 76 (38%) finished high school, and 19 (9.5%) had a university education. Fourteen persons declared themselves as unemployed. Of participants, 107 (53.5%) were still happy with the decision they had made to marry with another carrier person, 85 (42.5%) were not happy with their decision, and 8 (4%) did not answer the question. The reason for unhappiness was expressed as financial costs of PND in 47 (55.5%) (in other words they would be happy if they did not have to spend money on PND). Thirty-eight participants (44.5%) believed that they should not marry with another carrier person even if services are free of charge. In another question with the same content (used in a different center to reduce the bias), 110 (55%) persons answered that if they have knowledge of the problems in having healthy children, they would not marry another carrier. Ninety-three percent had a positive attitude towards the behavior of the counselors, however, 5% did not answer this question. Eleven percent of men had a high socioeconomic status according to the definition, 71% had average, and 81% had low. Fifty-seven percent of wives were defined as high regarding the socioeconomic class status, 38% had an average and 5% had poor. Concerning knowledge, 68.5% (number of couples/fertility rates) of participants had good and 30% had an acceptable knowledge. No statistically significant correlation was detected between knowledge and gender (p=0.97) or any other demographic parameters, except that the rural couples had a better knowledge than the urban couples (p=0.039). Couples with consanguine marriage had a better knowledge (p=0.0019). The attitude of absolutely relatively positive participants was 81.7% and relatively positive was 18.3%. Positive attitude was significantly correlated with good knowledge (p=0.009). No significant relationship was found between attitude and demographic parameters; including consanguine marriage. Those who had a positive attitude were not significantly regretful regarding marrying with another carrier person (this is in evaluating the relationship of total attitude toward one of the statement "what is your feeling on your marriage with a carrier?") (p=0.045). Regarding reproductive behavior practice (**Table 1 and Figure 1**), 50% of families had no markers for "unfavorable practice." While 27 (actual number) families had one marker (of unfavorable practice), 14 families had 2 markers, 8 families had 3 markers, 2 families had 3 markers, and 1% had 4 markers. A statistically significant risk factor for unfavorable practice was living in the city (p=0.019) (OR=1.61, 95% CI: 1-2.6). Unfavorable practice was not significantly related to lower knowledge scores (p<0.018). Unfavorable practice was not statistically different in consanguine versus unrelated marriage (p=0.29), and also was not related to regret concerning their decision (p=0.099). Families with unfavorable practice evaluated the behavior of counselors as comforting and appropriate, consanguine marriage was not statistically related to the attitude (p=0.49) or practice of families (p<0.29), but consanguine couples were more unhappy regarding the behavior of counselors (p=0.019). Socioeconomic status was not statistically related to knowledge (p=0.88), attitude (p=0.85) or practice (p=0.027) in both genders. One hundred and twenty pregnancies were recorded (28 couples had one pregnancy, 19 had 2, 14 had 3, and 3 had 4). Ten families for 2, and 11 families for 3 pregnancies did not accept PND. Four TM patients were born in the cohort. Two cases belonged to families, which had immigrated from other provinces with affected children.

Table 1 - Reproductive behavior of 100 at risk woman with thalassemia major children (1997-2006).

Reproductive behavior	n
Pregnancies	120
Unplanned	19
PND	26
Abortion with PND	6
Abortion without PND	7
Pregnancies without PND	68
Affected children	4

PND - prenatal diagnosis

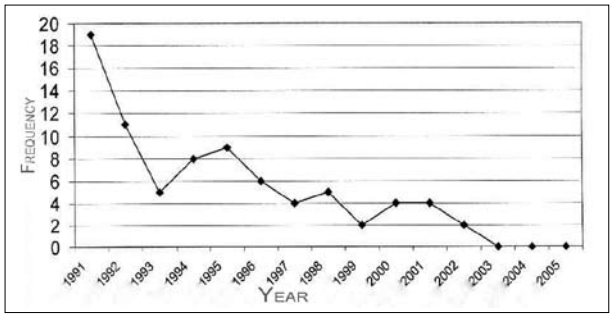

Figure 1 - Reproductive behaviors of 100 beta thalassemia carrier Iranian couples in 2006.

In 430 married couples with an incidence of having a child with serious genetic disease, 98 episodes (0.2 a year, 95% CI= -0.2-0.6) of unfavorable practice have been detected. The contraceptive method used by 12% of couples is unsafe. Eighty percent of couples used safe methods of contraception, and 8% were willing to have a well-planned pregnancy. The couples were counseled on the safe methods of contraception for an average of 6.5±5.8 (1-20) sessions, however, 26% cannot remember any participation in educational sessions. Financial problems was expressed as an important barrier for using PND in 60 persons, while 150 persons believed that it was not so. One half of the participants with or were aware of insurance coverage for PND expenses, 21% believed that there was no insurance support, and 28% said that they were not sure. Actual numbers of TM patients are shown in **Figures 2 & 3**. The total number of TM patients expected to be borne if no preventive program was in place was calculated (fertility percentage/carrier rate) as 25, as one can notice a 92% reduction in birth of TM patients achieved during the years of the prevention program.

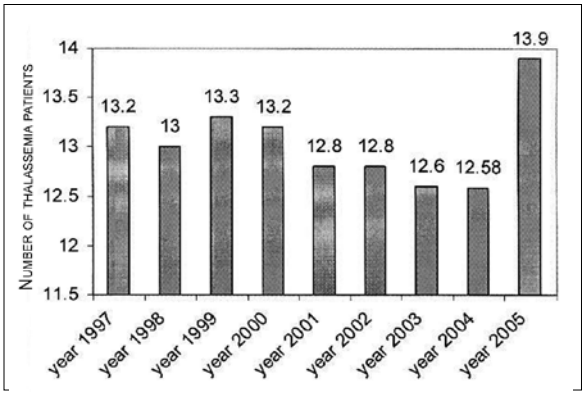

Figure 2 - Birth rate/1000 in the province (birth rate of TM patients in the general population).

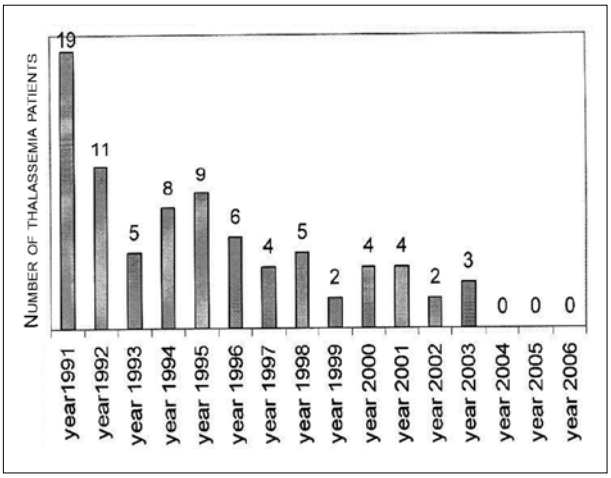

Figure 3 - Actual new thalassemia major registered patients with the same health center where 240 at risk couples have been registered since 1997, and after starting screening and counselling.

Discussion. High risk couples, 14 years after starting counseling still have good knowledge of TM regarding its genetic nature, risk of a child being affected at each pregnancy, ways of prevention, and treatment. This finding can be explained by repeated counseling as well as transport of information through the media.[3,4] They also had a strong positive attitude toward prevention, and planning . In a cross-sectional study of Hajian,[8] 536 persons in a similar population showed that approximately 81% of men and 86% of women who were interviewed knew about thalassemia. The source of information was radio and television in the majority (64%) and knowledge and attitude were better in residents of an urban area. The socioeconomic status of wives was better than husbands as before. Higher scores were recorded in females if they have a university degree, or if they have a job with any amount of income. As a result, almost 70% of subjects believed that financial problems was not the main barrier for using PND services, hence, 30% were not able to pay the costs of PND. This concern was responsible for the "regretfulness" of their decision. At that time, costs of molecular studies for couples and for PND were not covered by health insurance companies. Fortunately, this problem has been solved in 2007 and costs for transportations of couples are being paid if needed. In least 430 married couple, 98 episodes of inappropriate behaviors happened and ended to the birth of 4 TM patients. Sixty-eight pregnancies ended without PND, which is less than anticipated according to the one in 4 chance for autosomal recessive inheritance, which is 24 affected offspring. The reason is that in these families either one or both spouses are suspicious for being a beta thalassemia gene carrier (microcytosis and hyporchromia with normal hemoglobin A2). Laboratory facilities for alpha-thalassemia detection were not available at that time. Nowadays, most of these cases proved to be alpha-thalassemia carriers.[9]

Regarding social and educational class of this population, is it a failure or success? In an unpublished study, we noticed that families who had TM patients, the time interval between pregnancies was shorter than families without any affected children. The number of pregnancies and illegal abortions also were higher. There are arguments regarding genetic counseling in 2 ways. One was concerning informed decisions by couples and not forcing them to separate, which is what is more or less practiced in different centers.[2,4,10,11] The other concern, which is expressed by other people is to prevent couples from marrying another carrier to avoid increasing gene

frequency.[12,13] We would like to emphasize that the goal of community genetic services is to help at risk couples to have healthy children, not to decrease the gene frequency. Therefore, the best way is to offer complete information and different options to couples, and let them to choose what is best for them.[10,11]

In 2002, Petrou et al[10] published the productive behavioral of 102 couples at risk for TM affected children, all selected by screening and finished their reproduction period (age of wife >42). They were mainly former Cypriot (70%), Indian, Pakistani and Bangladeshi UK residents.[12] Their definition of "favorable pregnancy" was different with ours, as having an affected child as an informed choice was regarded as "unfavorable practice" in our study, whereas this was defined as favorable in their study. We also considered unplanned pregnancy as "unfavorable practice," but they did not, and it seems that we were "very strict" in our study. They reported that when early losses are included, 58% of pregnancies had a favorable outcome, but only 30% of couples had only favorable outcomes, and 41% lost 2 or more pregnancies. Though, 88% of prospectively detected couples achieved a thalassemia-free family, the findings leave no room for complacency. Forty-three percent of the couples were lucky and had only unaffected live births. It might be argued that these couples derived no benefit from screening, but suffered unnecessary stress. In the past, they did not need prenatal diagnosis. However, the data shows that these couples used their early knowledge of risk to maximize their chance of having a healthy family, by strictly limiting reproduction after a minimum acceptable number of unaffected children.[10]

How can the program of counseling and prevention possibly be more successful? More and periodic instruction programs theoretically will help, for example: families who use unsafe contraceptive methods, had unplanned pregnancies or failed to use PND services must come under more intensive supervision follow-up. The best media for mass education is television, and a recent survey by Khorasani et al[3] showed that 98% of families have a TV set. Also, there is a positive attitude toward educational material, offered via TV programs (in Farsi). Our study population was limited and a study in larger populations, and other cultures and countries with at risk populations is recommended.

In conclusion, thalassemia prevention program improved the general knowledge, attitude, and practice of high-risk couples and carriers.

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