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Original Article

Attitudes of pregnant women and mothers of children with orofacial clefts toward prenatal diagnosis of nonsyndromic orofacial clefts in a semiurban set-up in India

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ABSTRACT

Objectives: To assess the attitudes of pregnant women and mothers of children with orofacial clefts toward prenatal diagnosis of clefts and elective termination of pregnancy, and to investigate their opinion about who makes reproductive decisions in the family. **Design:** Two hundred subjects were included in the study prospectively regarding hypothetical prenatal ultrasound diagnosis of clefts. **Setting:** The study was done in a private tertiary care institution and a teaching hospital. **Subjects/Participants:** One hundred pregnant women consulting the Obstetrics department and 100 mothers of children with orofacial clefts in the Cleft and Craniofacial Unit were selected. **Materials and Methods:** Group I subjects were interviewed using a questionnaire and were shown preoperative and postoperative pictures of children treated for cleft lip and palate. Group II subjects were interviewed using a questionnaire. **Results:** Only 3% of Group I subjects and 2% of Group II opined that they would choose the elective termination of pregnancy if the fetus was diagnosed with a cleft on an ultrasound scan. In Group II, 70% subjects wished to have known about pregnancy affected with cleft prenatally and 96% said they would definitely avail ultrasound scans to determine pregnancy affected by clefts in future. **Conclusions:** Majority of the respondents from both the groups chose to continue with the pregnancy affected with a cleft when questioned regarding hypothetical prenatal ultrasound diagnosis of the cleft.

KEY WORDS

Orofacial clefts, prenatal ultrasound diagnosis, termination of pregnancy

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INTRODUCTION

Facial deformity, especially around the mouth, is rated as the least attractive of all deformities.[1,2] Orofacial cleft is one such condition which affects lip, alveolus, and hard and soft palate either unilaterally or bilaterally. Though comprehensive treatment for an

orofacial cleft is now available universally, an increased awareness of cosmetic appearance in society and the availability of prenatal ultrasound diagnosis have led to evidence of elective termination of pregnancy.[3,4] Since non-syndromic orofacial clefts are nonlethal birth defects, prenatal ultrasound diagnosis and subsequent termination of pregnancy is an important ethical issue.[5,6]

Transvaginal sonography due to its high resolution and proximity to the fetus can detect 95% of fetal malformations at 14–16 weeks of gestation.[7-10] The detection of orofacial clefts is being routinely included in ultrasound screening practices in some countries[11,12] and providing reproductive choices to pregnant women is one of the objectives of such fetal anomaly screening programmes.[13] The availability of this advanced technology in the Indian subcontinent can offer reproductive choices to pregnant women who were hitherto unexposed to the prenatal diagnosis of clefts.[14] The elective termination of pregnancy for prenatal diagnosis of a non-syndromic oral cleft has been viewed differently across the world, the concerns being ethical, religious, and cultural differences among the population.[15,16]

The incidence of terminations of pregnancy for non-syndromic facial clefts because of prenatal diagnosis ranges from 0% to 92 %[17] whereas the rate of termination is high when the cleft is associated with other malformations.[18-20] The attitude of the Indian population toward individuals affected by clefts is not discriminative as the acceptance of cosmetically deprived patients is high in contrast to the population of developed countries where the practice of elective termination of pregnancy is prevalent.[4] To our knowledge, no previous studies have been conducted in the Indian subcontinent to assess the perceptions about the prenatal ultrasound diagnosis of orofacial clefts and the option of elective termination of pregnancy in our population. A study was designed to assess the attitudes of pregnant women and mothers of children with orofacial clefts toward a hypothetical situation of prenatal ultrasound diagnosis of clefts and elective termination of pregnancy and passed through our ethical committee. Though hypothetical, this study is important to determine future treatment needs in our country where every year 30,000 babies are born with orofacial clefts.[21]

MATERIALS AND METHODS

Our center is a tertiary teaching hospital with 2000 beds catering to the needs of a population of 1.5 million. The Cleft and Craniofacial Unit was established in year 2001 and since then 5000 patients with cleft lip and palate have been managed successfully. It consists of a multidisciplinary team of a cleft surgeon, orthodontist, pediatrician, anesthesiologist, speech therapist, trained nurse, and social worker. The study consisted of 200 patients who were divided into 2 groups. Pregnant women reporting to the Obstetrics outpatient department of the hospital were randomly assigned to Group I and inclusion criteria for Group I was all pregnant women in the first and second trimesters. Group II consisted of mothers of children with an orofacial cleft reporting to the Cleft and Craniofacial Unit. These subjects were randomly enrolled in the study between November 2009 and May 2010. Inclusion criteria for subjects in Group II were mothers of children primarily operated for cleft lip, cleft palate, and cleft lip and palate both. All the subjects were interviewed in the outpatient department of Obstetrics and Cleft and Craniofacial Unit for Groups I and II, respectively, using a structured questionnaire and also demographic information by a single interviewer in subjects' vernacular language. Informed consent was taken from all the subjects and they were encouraged to answer the questions and were also counseled regarding the etiology and treatment of clefts. Ethical clearance was obtained from institution's Ethical Council.

The questionnaire devised was intended to collect the information regarding the literacy level, occupation of spouse, caste and religion, and whether they live in a joint family or nuclear family. The intention was to acquire data that might influence decision making when confronted with the situation of having a child affected with cleft lip palate. The study was piloted earlier on 10 subjects from both the groups so that ambiguous questions were modified.

RESULTS

Table 1 represents the demographic data of the subjects enrolled in Groups I and II of the study. In Group I, 82% of subjects were less than 25 years of age, 87% were educated, being high school graduated or more, 92% were housewives, 75% of subjects' husbands were self-employed in a small business, and 21% were farmers implying that almost 96% belonged to the lower socioeconomic stratum; 95% of the subjects studied were Hindu and 85% of them lived in joint families.

In Group II, 76% of children with cleft lip palate were under 2 years of age; 96% were diagnosed as non-syndromic oral

Table 1: Sociodemographic data of Group I and Group II respondents

Characteristic	*Group I*	*Group II*
Respondents' age (years)		
18–20	24	
21–25	56	
26–30	15	
31–35	5	
>35	0	
Education		
≤7th grade	7	20
8th–10th grade	54	25
11th12th grade	25	9
Graduate	9	10
Illiterate	5	36
Occupation		
Housewife	92	73
Employed	8	27
Spouse's occupation		
Agriculture	21	44
Self-employed	75	16
Professional	4	40
Religion		
Hindu	95	89
Muslim	5	10
Christian		1
Type of family		
Joint family	85	69
Nuclear family	15	31
Area of residence		
Rural	60	66
Semiurban	40	34

clefts. Seventy-four percent of respondents were post-high school educated; 44% were farmers and 40% employed as laborers. A total of 89% were Hindus; 73% of mothers were housewives and 69% of them lived in joint families. Hundred percent of the subjects in both the groups were from rural and semiurban areas.

Table 2 presents the responses of the subjects to the hypothetical question on their attitude toward the prenatal ultrasound diagnosis of cleft lip and palate. Three percent subjects in Group I and 2% in Group II chose the option of medical termination of pregnancy. A total of 70% of Group II subjects wished they had known about the prenatal diagnosis of cleft and 71% expressed that prenatal ultrasound diagnosis is important. Only 1% in Group I and 6% in Group II subjects expressed that they would be the sole decision maker in the family for reproductive choices in the context of pregnancy detected with a cleft fetus.

DISCUSSION

With the increasing awareness regarding the availability of the comprehensive treatment for cleft lip and palate in

Table 2: Responses of Group I and Group II

Question	*Group I*	*Group II*
Choice of reproduction if prenatal ultrasonography revealed a fetus with a cleft		
Continue pregnancy	94	96
Medical termination of pregnancy	3	2
Refused to answer	3	2
Importance of knowing the diagnosis of a cleft prenatally		
Extremely important	62	24
Important	29	51
Somewhat important	9	20
Not important	0	5
Decision maker in the family		
Respondent herself	1	6
Father of the child	37	35
Elders/in-laws at home	34	37
Combined (all together)	20	2
Father and mother of the child	5	20
Refused to answer	3	0
Had known diagnosis prenatally		
Yes		9
No		91
Wished to have known prenatally		
Yes		70
No		13
Not sure		17

developing countries due to better access to health care and a desire to have an aesthetically perfect child, various non-governmental organizations have been successful in rendering free cleft services in these countries.[22]

The rationale for including mothers in the study was to assess the attitudes of women in particular in both groups toward the prenatal diagnosis of clefts and their opinion about who will be the decision maker in the context of the prenatal ultrasound diagnosis of a cleft. Studies have shown that the role of women in decision making in pregnancy and abortion was undersized[23] and their husbands and in-laws had a greater role in decision making.[24] In this study, when pregnant women and mothers of children with a cleft were hypothetically questioned regarding their choice of reproductive option if prenatally diagnosed to have a cleft fetus, only 3% of pregnant women and 2% of mothers of children with a cleft chose to electively terminate the pregnancy.

A study by Bronshtein and Blumenfeld[25] in Israel showed that 14 out of 15 patients voluntarily terminated pregnancy when a cleft was diagnosed by prenatal ultrasound. This is notable in that all the 15 couples had consulted parents of children with a cleft who had undergone surgical repair. In the follow-up of the same

study, authors[26] quoted out that of 24 patients diagnosed for the cleft, 23 chose to terminate the pregnancy. In our study, the subjects were shown pre and postoperative pictures of the child with cleft lip palate, and the responses of the pregnant women were to a hypothetical question that in case their ultrasound scan diagnosed a cleft, what reproductive choice they would make. However, in the published studies,[25,26] the subjects were pregnant women who were actually diagnosed with fetal cleft lip and palate by ultrasound. The responses might be subjective for the simple reason that the decision varies when one actually has to face a situation than when one hypothetically reacts to a situation.

The responses of mothers of children with clefts to the same question are similar to the results of a study conducted in Argentina by Wyszynski, Perandones, and Bennum[16] wherein most parents of children with a cleft strongly support the continuation of pregnancy as they felt that an oral cleft is not a serious condition requiring the termination of pregnancy and only 6.4% of 165 parents chose to terminate pregnancy. The reason for the continuation of pregnancy with a cleft child in Argentina is explained on the basis that most subjects in the study were Catholics and abortion is legally restricted in Argentina. The reasons for similar results in our study may be attributed to cultural and religious beliefs in this region.[27]

In both the groups, the main decision maker in the family was found to be either the husband of the woman or the elders/in-laws at home. Though these women are educated and aware of the cleft condition, majority of them live in joint families where the head of the family makes the decisions. An extensive study conducted by Alok Ranjan[28] on the institutional context of fertility and reproductive health in Madhya Pradesh, India, highlights the institutional environment – the family, the society, the culture and tradition, etc. – in which people have strong bearings on reproductive health decision making and hence on fertility and reproductive health outcomes.

Majority of the subjects in both groups of our study felt that it was important for them to know and become aware of conditions like cleft prenatally. Some subjects opined that being aware of conditions prior to child birth makes one capable of handling such situation better when actually confronted. Few of the subjects in Group II expressed the concern that though they underwent ultrasound scans during pregnancy, the obstetrician might not have informed them about the diagnosis prior to birth.

Several studies have revealed that prenatal counseling about the cleft is thought to give the family an opportunity to learn about clefts and become prepared for the event and our study strongly supports the importance of prenatal diagnosis. Parents may be counseled about how the immediate needs of their newborn will differ from infants born without an orofacial cleft. Parents will have time to prepare by talking with health care providers, family members, and tapping multiple resources to learn more about orofacial clefts.[29-33]

In contrast, some questions remain as to whether the early knowledge of a pregnancy with a cleft child may generate greater maternal anxiety during a presumably joyful period, whether prenatal information is of any value because the problem cannot be corrected prenatally,[34] and whether prenatal diagnosis might encourage the termination of the pregnancy in the absence of other malformations.[35] However, it emphasizes the need of prenatal counseling to pregnant women subsequent to prenatal diagnosis of cleft lip and palate.

The differences noted in our study can be contributed to many factors, the most important one being the eastern family values like more emphasis on deeds than appearance and acceptance of physically deprived individuals and the law of *karma*.[36] It may also be attributed to facts like lower education and awareness level, and lack of advanced health care access and reproductive choices.[37] We did not compare the two groups in our study as the groups are not matched and hence not comparable but in spite of these demographic differences, it is noteworthy that the attitude of two sets of mothers toward the prenatally diagnosed cleft baby remains essentially the same which goes to suggest the general attitude of our society. Our study is the representative of a particular socioeconomic group. A larger sample size of a different socioeconomic class is required to assess the attitudes toward the prenatal ultrasound determination of pregnancy with a cleft child. It would be prudent to conduct studies in actual pregnant women diagnosed with a cleft fetus.

The attitude of pregnant women and mothers of children with clefts would seem to indicate that, unlike studies in developed nations, the trend of accepting a child with an abvious congenital anomaly is far higher in our society. And this tolerance is expected in a civilization as ancient and as enriched as ours, which has always cherished deeds and values and treated appearance as rather trivial in comparison.

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