

# **Attitudes toward reproduction by parents of children with nonsyndromic oral clefts in Argentina**

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## **Abstract**

### **Objective**

To investigate the attitudes of Argentine parents of children with nonsyndromic oral clefts with respect to procreation and to assess the variables that might influence their responses.

### **Design**

One hundred and sixty five parents of children with oral clefts ascertained from a craniofacial clinic in the suburbs of Buenos Aires answered a 151-item semistructured questionnaire. The questionnaire included sections covering sociodemographic information, level of religiousness, characteristics of the child's cleft, parental perception of their child's cleft, pregnancy history, recurrence risk, access to health care, attitudes in regard to abortion, and family environment.

### **Results**

Most parents (65.1%) believe their child's cleft is not a serious condition. None of the respondents would terminate a pregnancy because the ultrasound reveals an oral cleft. Similarly, very few (6.4%) would terminate the pregnancy if there is an early diagnosis of Down's syndrome. Half of the respondents believe that abortion should not be an option for any couple expecting a child.

### **Conclusions**

Most respondents do not perceive oral clefts as a severe condition. Parents would not choose to terminate the pregnancy over delivery of such an affected newborn.

Keywords: cleft lip and palate; recurrence risk; pregnancy termination; genetic counseling; psychosocial.

## Introduction

For most of the severe genetic diseases, prevention is based during pregnancy on prenatal diagnosis and termination of the pregnancy of affected fetuses (Zlotogora, 2002). As prenatal diagnostic technologies become more sophisticated and available, they are being used increasingly more frequently for nonlethal birth defects that are mildly or little incapacitating. On the basis of the prenatal diagnostic findings, parents may then choose to continue the pregnancy with new awareness and armed with information, may seek prenatal therapy if that is available, or they may choose to terminate the pregnancy (Strauss, 2002).

The first published report of prenatal diagnoses of oral clefts (OC) by real-time ultrasonography dates to 1981 (Christ and Meininger, 1981). Twenty years later, and despite improved scanning protocols, the detection rates are still low, ranging from 30% (Levi et al., 1995; Davalbhakta and Hall, 2000) to 48% (Boyd et al., 1998). The Eurofetus study (Grandjean et al., 1999) of routine ultrasonography in pregnancy showed a detection rate for cleft lip of 25%, for cleft lip and cleft palate of 22%, and for isolated cleft palate of only 1.4%. The more recent use of three-dimensional ultrasound (3D US) promises to dramatically increase these identification rates (Dyson et al., 2000; Chmait et al., 2002; Wayne et al., 2002).

Since nonsyndromic OC (NSOC) are nonlethal birth defects, the use of prenatal diagnosis raises important ethical issues (Blumenfeld et al., 1999; Hager, 2002). This prompted the American Cleft Palate-Craniofacial Association to hold a panel discussion entitled "Beyond easy answers: Prenatal diagnosis and counseling during pregnancy" during its 2000 annual meeting (Strauss, 2002). While the theoretical framework for prenatal diagnosis and counseling has been laid out (see the review of Jones, 2002), few articles have addressed parental choices with respect to NSOC.

In Argentina, as in all other Latin American countries except Cuba, Guyana, and Puerto Rico, abortion is legally restricted. The Penal Code of Argentina classifies abortion as a crime against life and against the person, and condemns to imprisonment both the person who carries it out and the woman who self-induces it or agrees to have it performed (Gogna et al., 2002). The Code establishes only two exceptions: (1) if the woman's life or health is at risk and there is no other way of saving her or preserving her health; or (2) if the pregnancy is the result of a "mentally insane or idiot woman" having been raped. Despite these restrictions, close to half a million abortions are performed in Argentina annually, most in terrible conditions (Carbajal, 2003). The purpose of this study was to investigate the attitudes of Argentine parents of children with NSOC with respect to procreation and to assess the variables that might influence their responses.

## Sample and Methods

The Asociación Piel is a large outpatient craniofacial clinic in Avellaneda, an industrial suburb of the city of Buenos Aires. It provides multidisciplinary assessment and treatment services, including plastic surgery, pediatrics, dentistry, speech and language pathology, ear/nose/throat, psychology, and genetic counseling to 141 new patients and 482 follow-up patients with craniofacial conditions per year. The study sample consisted of 165 parents of children with NSOC who were treated at Asociación Piel between June 2000 and August 2001 and who agreed to answer the questionnaire at one of the clinic visits. Sixty-three percent of the respondents were mothers, 27.8% were fathers, and close to 9% were other family members. Seventy-two percent of respondents resided in the province of Buenos Aires, while close to 17% were from the city of Buenos Aires. A trained study researcher was always available nearby to answer any questions with regards to the questionnaire. As part of the inform consent process, all participants were encouraged to answer the questions as truthfully as possible. Since abortion is legally restricted in Argentina, respondents were assured strict confidentiality. The study was approved by the local Ethics Committee.

Data were collected using a 151-item semistructured questionnaire. The questionnaire included sections covering sociodemographic information, level of religiousness, characteristics of the child's cleft, parental perception with respect to their child's cleft, pregnancy history, recurrence risk, access to health care, attitudes in regard to abortion, and family environment. Most questions were of yes/no type, multiple choice, and open-ended. For some items, parents responded on a 10-point Likert scale.

The survey was developed and administered to 10 mothers of children with NSOC for pilot testing. This was particularly important given, to the best of the authors' knowledge, this

type of survey has not been used in Argentina before. Questions that were thought to be confusing or ambiguous were modified as needed. Statistical analyses were carried-out with the software Stata, version 7.0 (Stata Corporation, 2001).

## Results

Table 1 presents demographic characteristics of the population under study. Of the 165 respondents, most were mothers (63.0%), married (76.1%), living in non-rural areas (99.4%) and employed (64.6%). Close to half of them (42.1%) had post-high school education and the vast majority (90.3%) were Roman Catholic. Eight percent of the children had either a unilateral or a bilateral cleft lip with a cleft palate.

Table 2 includes parents' perceptions on the severity of their children's NSOC and on the importance of prenatal diagnosis for NSOC. After adapting some of the questions used by Gooding et al. (2002) for achondroplasia, participants were asked to place nonsyndromic oral clefts on a spectrum ranging from trait (1) to disorder (10). The mean response ( $\pm$  standard deviation) was found to be  $3.3 \pm 2.4$ . Participants were then asked to choose one word from a list to describe nonsyndromic oral clefts. The distribution of responses was bimodal, with 37.3% considering it a "trait" and 30.7% calling it a "disorder". By asking "Is your child's cleft a serious or a not serious condition?" (see Table 2 for exact wording), an effort was made to obtain a subjective assessment of how parents felt their children were clinically affected. In the study, 53 (34.8%) subjects felt their children were severely affected. No significant correlation was found between the medical classification of the NSOC and the parents' own perception of severity. This apparent discordance has been seen in other studies measuring subjective assessment of "burden" attached to a disorder (Pearn, 1973; Benjamin et al., 1993). Only 4% of the parents knew during pregnancy that the child had a NSOC. When asked whether they wished they had known about it, 65.1% responded positively, with 53.1% considering prenatal diagnosis to be "extremely important". When asked whether they would seek an ultrasound to identify a cleft in a future pregnancy, 78.8% answered that they would do it.

None of the respondents would terminate a pregnancy because the ultrasound reveals a NSOC. Similarly, very few (10 parents or 6.4%) would terminate the pregnancy if there were an early diagnosis of Down's syndrome. Half of the respondents believe that abortion should not be an option for any couple expecting a child. When asked to assume a 5% risk of having another child with a cleft, the answers were equally divided between those who would request prenatal diagnosis in early pregnancy and those who would not solicit it at all.

## Discussion

The findings of this study indicate that parents of children with NSOC in Argentina believe that this isolated treatable birth defect is not a reason to terminate a pregnancy. Most parents believe elective termination of pregnancy should not be offered as an option in general and that they would not interrupt a pregnancy if the fetus were diagnosed prenatally with Down's syndrome. Though parents stated it would not change their decision to continue the pregnancy, most wanted thorough, honest information to adequately prepare for parenting a child with a cleft.

As mentioned in the Introduction, few articles have addressed parental choices with respect to oral clefts. Andrews-Casal et al. (1997) obtained questionnaire information from 61 families who had at least one child with an oral cleft. The majority of mothers and fathers indicated that their reproductive plans had not changed because of the birth of their child with cleft lip with or without cleft palate (CL/P). Bronshtein et al. (1996) detected 15 cases with CL/P during a 10-year period and over 24,000 early (14 to 16 weeks gestation) transvaginal sonographies in northern Israel. Fourteen of the 15 pregnancies were voluntarily terminated, after the couples consulted other parents of children who had been born with CL/P and had undergone surgical plastic repair. Also, in a small survey of 17 families of children with oral clefts, all parents said that they would choose termination in a subsequent gestation if cleft lip was detected. In a follow-up, these authors (Blumenfeld et al., 1999) increased the numbers to 24 cases of CL/P identified using transvaginal diagnosis, of which 23 (95.8%) chose to terminate the pregnancy. Analyzing data from the Hawaii Birth Defects Program, Forrester et al. (1998) identified 32 prenatally diagnosed cases with CL/P. Of those, 9 (28.1%) were terminated. Jones (1999) reported that of 8 cases with apparently isolated CL/P identified before 22 weeks gestation, 6 women continued the pregnancy and 2 (25%) did not. Davalbhakta and Hall (2000) stated that of 30 British patients diagnosed with an oral cleft with

early sonography, 3 cases (10%) terminated their pregnancy. Two of these cases had multiple other abnormalities including holoprosencephaly. Only one woman actually terminated her pregnancy due solely to the antenatal diagnosis of a bilateral cleft lip and palate.

The striking differences observed when comparing these studies to ours might be explained by the combination of a variety of factors: 1) religious, since close to 90% of the respondents to our questionnaire consider themselves practicing members of the Catholic faith; 2) legal, since abortion is legally restricted in Argentina and the conditions in which they are performed are extremely poor; 3) social, because abortion produces stigma and isolation; 4) financial, since abortions are largely illegal, women have to pay it in full out of their own pocket; 5) perception of burden of a cleft, because the majority of our respondents believe that an oral cleft is not a serious condition.

Our study has some methodological shortcomings that must be mentioned. First, the subjects were ascertained from a single craniofacial center in the suburbs of the city of Buenos Aires. It is possible, although unlikely, that there is an unintentional ascertainment bias, wherein study participants do not represent the entire population of families of children with NSOC in this country. The demographic distribution of the survey respondents, however, closely resembles that of the general population in Argentina. It should also be mentioned that in this clinic parents receive genetic counseling by an experienced craniofacial geneticist. This might have influenced some responses, as several parents had received genetic counseling on an occasion before answering the questionnaire. Second, due to financial constraints, no “parent controls” (ie, parents of children without birth defects) were collected as part of this study. A second phase of this study contemplates a follow-up of the participants from the original cohort and the recruitment of parent controls. Third, the responses are hypothetical decisions in an artificial setting (ie, identification in early pregnancy of a fetus with an oral cleft or with Down’s

syndrome). Research on offering genetic testing both for individual risk (ie, Huntington's disease and cancer susceptibility) and prenatal diagnosis (ie, cystic fibrosis) indicates that interest in using genetic testing is often greater than actual use of such tests (Wiggins et al., 1992; Jedlicka-Kohler et al., 1994; Biesecker et al., 2000). A follow-up study might be helpful in identifying what parents really do under these circumstances, although it is likely that the sample size would be too small to draw meaningful conclusions. Fourth, the results of this survey might not be generalizable to other populations, since cultural, religious, and socioeconomic factors are known to influence parental reproductive choices and their willingness to accept the birth of a "less than perfect" child (Jones, 1999). Finally, since abortion is largely illegal in Argentina, some parents might have felt compelled to answer that they would not consider having one out of fear of identification or possible stigma. However, all of the subjects had children between 1 and 29 years old who were being treated at the clinic and had developed a relationship of trust with its professionals. We believe this relationship increased their level of comfort in responding honestly to the questionnaire; however, we do not negate the potential risk to validity from a socio-scientific perspective.

This study is the first to describe reproductive attitudes of families of children with NSOC in Argentina and one of few published worldwide. We conclude that parents in Argentina do not perceive oral clefts as severe conditions and that they would not choose to terminate the pregnancy over delivery of such an affected newborn.

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**Table 1. Characteristics of respondents\*.**

<b>Characteristic</b>	<b>N = 165</b>
Respondent's Age (mean, SD)	36.1 (9.5)
Child's Age (mean, SD)	6.4 (4.8)
Mother Respondent	104 (63.0)
Area of Residence	
<i>Rural</i>	1 (0.6)
Small city/town	73 (44.8)
Major metropolitan area	89 (54.6)
Marital Status	
<i>Single/Never been married</i>	20 (12.3)
Married	124 (76.1)
Divorced/Separated	15 (9.2)
Widowed	4 (2.5)
Employment	
<i>Full-time</i>	50 (32.3)
Part-time	50 (32.3)
Unemployed	26 (16.8)
Retired	1 (0.7)
Highest School Level Achieved	
<i>Less than high school</i>	46 (28.1)
High school diploma	49 (29.9)
<i>Some college or technical school</i>	18 (11.0)
<i>College diploma</i>	23 (14.0)
<i>Some graduate school</i>	12 (7.3)
<i>Graduate diploma</i>	16 (9.8)
Roman Catholic Religion	149 (90.3)
Attends Religious Services	
<i>Several times a week</i>	6 (3.6)
<i>Once a week</i>	14 (8.5)
<i>Two to three times a month</i>	15 (9.1)
<i>About once a month</i>	14 (8.5)
<i>Several times a year</i>	48 (29.1)
<i>Once or twice a year</i>	46 (27.9)
<i>Never</i>	22 (13.3)

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Type of nonsyndromic oral cleft child has	
<i>Unilateral cleft lip without cleft palate</i>	11 (6.7)
<i>Unilateral cleft lip with cleft palate</i>	69 (41.8)
<i>Bilateral cleft lip without cleft palate</i>	6 (3.6)
<i>Bilateral cleft lip with cleft palate</i>	63 (38.2)
<i>Cleft palate only</i>	16 (9.7)

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\*Expressed as *n* and (%).

**Table 2. Parents' perceptions about their children's oral cleft and prenatal diagnosis\*.**

	<b>N = 165</b>
"People have a range of attitudes about various traits/conditions. With trait (such as hair color or eye color) on one end of the spectrum and disorder on the other, where, in your opinion, does the cleft of your child fall?" (scale from 1=trait to 10=disorder)**	3.3 (2.4)
"If you had to choose one word, which of the following would best describe the cleft lip/palate of your child?"	
<i>Trait</i>	56 (37.3)
<i>Difference</i>	27 (18.0)
<i>Condition</i>	12 (8.0)
<i>Disability</i>	2 (1.3)
<i>Disorder</i>	46 (30.7)
<i>Disease</i>	7 (4.7)
"Which statement best reflects your view about your child's cleft lip/palate?"	
<i>Your child's clefts is not a serious condition</i>	99 (65.1)
<i>Your child's clefts is a serious condition</i>	53 (34.8)
"Did you know that your child had cleft lip/palate before he/she was born (for example, through ultrasound)?"	
<i>Yes</i>	6 (4.0)
<i>No</i>	145 (96.0)
"Do you wish you had known that your child had a cleft lip/palate before he/she was born?"	
<i>Yes</i>	95 (65.1)
<i>No</i>	17 (11.6)
<i>Not sure</i>	34 (23.3)
"In your opinion, how important would it be to know before birth if the child has a cleft lip/palate?"	
<i>Extremely important</i>	78 (53.1)
<i>Important</i>	43 (29.3)
<i>Somewhat important</i>	15 (10.2)
<i>Not at all important</i>	11 (7.5)
"Do you think that you would request an ultrasound to determine whether you have a child with a cleft in a future pregnancy?"	
<i>Yes</i>	119 (78.8)
<i>No</i>	32 (21.2)

\*Expressed as *n* and (%). \*\*Expressed as mean and standard deviation.

**Table 3. Parents' responses regarding reproductive choices\*.**

	<b>N = 165</b>
"Would you ever consider an abortion because the child has a cleft lip/palate?"	
Yes	0
No	153 (97.4)
Unsure	4 (2.6)
"Would you ever consider an abortion if the child had Down's syndrome?"	
Yes	10 (6.4)
No	107 (68.2)
Unsure	40 (25.5)
"In general, do you think that abortion should be an option for any couple expecting a child?"	
Yes	38 (24.1)
No	80 (50.6)
Unsure	40 (25.3)
"If you knew that your risk of having another child with cleft lip/palate was 5%, which of the following decisions would you make?"	
<i>I would certainly have more children</i>	22 (15.5)
<i>I would probably have more children</i>	15 (10.6)
<i>I would certainly have more children but I would request prenatal diagnosis</i>	25 (17.6)
<i>I would probably have more children but I would request prenatal diagnosis</i>	12 (8.5)
<i>I would certainly have more children but I would not request prenatal diagnosis</i>	0
<i>I would probably have more children but I would not request prenatal diagnosis</i>	4 (2.8)
<i>I am not able to have more children</i>	25 (17.6)
<i>I don't know</i>	39 (27.5)

\*Expressed as n and (%).

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