RELATIONSHIP OF PARENTAL HEALTH AND SOCIAL SUPPORT DURING PREGNANCY WITH THE RISK OF NON-SYNDROMIC ORAL CLEFTS: A CASE – CONTROL STUDY

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Abstract

Oral clefts are divided into cleft lip, cleft palate and cleft lip along with cleft palate. Cleft palate is defined as an inability of palatal shelves to approximate and close during the first months of embryogenic period thus leaving an opening in the roof of the mouth and forming communication between the nasal and oral cavities.

Objective: To determine the relationship of risk of oral clefts with parental health and social support.

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Study Design: Case Control Study.

Study Setting and Duration: Various hospitals in Lahore and 3 month duration.

Materials and Methods: A Case Control Study was conducted. A total of 100 subjects (50 cases, 50 controls) were included. Sampling was done by purposive method. Data was collected with the help of Cornell Medical Index Health Questionnaire and Social Support Questionnaire after taking their informed consent. Data was analyzed by using SPSS version 20.0. Mean and standard deviation were used to describe quantitative variables like age. Test of significance were applied.

Results: 56% of cases were females and 44% were males. Mean age of mothers was 27 and fathers was 31. Most mothers were uneducated (36%) while most fathers were educated till matric (34%). Income of most parents was in the range of 10,000-20,000 PKR (44%). 24% of controls were females and 76% were males. The associations of oral cleft was found significant with parental respiratory, cardiovascular, gastro-intestinal, musculoskeletal, skin-related, genitourinary health, easy fatigability, health habits, anxiety, anger, paternal nervous system health, sensitivity, and maternal frequency of illness, depression and feeling of inadequacy (p < 0.05). Maternal social support categories indicated that cases were less supported than controls: number of persons available and level of satisfaction (p < 0.05).

Conclusion: Oral clefts have a multi-factorial etiology. Hence, an effective preventive program should take into account parental health and social support factors.

Key Words: Non-syndromic Oral Clefts, Parental Health, Social Support.

Introduction

This is a case – control study to establish the relationship of mental and physical health of both parents and social support both formally and informally by the family and community during pregnancy with non-syndromic oral clefts.

Oral clefts are divided into cleft lip, cleft palate and cleft lip along with cleft palate. Cleft palate is defined as an inability of palatal shelves to approximate and close during the first months of embryogenic period thus leaving an opening in the roof of the mouth and forming communication between the nasal and oral cavities.[1] Cleft lip results due to failure of fus-ion of both sides of fetal lip tissue by 35 days of gestation. The defect may affect one side or both.[2] Esthetic defects which are associated with cleft lip and cleft palate in infants results in feeding problems, infections of ear, hearing impairement, dentition problems, difficulty in articulation, social, psychological and behavioral problems.[3]

The incidence of cleft lip in Pakistan is one in 500 live births. This is fourth largest in the world after China, India and Indonesia. Every day 25 cases in Pakistan are born with this deformity and in a year 9000 cases are added.[4] Many causative factors are found to be linked with cleft lip and palate. Researchers have majorly outlined risk factors like parental age,[5-7] family history,[8] consanguineous marriages,[9,10] co-morbidities like infections during pregnancy,[11] multigravida[12] in mothers of low socio-economic status, smoking,[13] alcohol,[14] drugs[15] (anti-epileptics, steroids, anti-neoplastics), occupational and environmental exposures.[16]

A research conducted in China studied oral clefts in relation to parental health and social support during pregnancy.[17] However, no such study has been conducted in Pakistan.

Our study will outline the parental health and social support factors associated with the said defect. This can help shift the paradigm of approach for prevention of this health problem towards a more holistically oriented one. Moreover, various fruitful research efforts can be based on the results of this research to ascertain the risk factors in case of health of both parents and support of family and society.

Materials and Methods

The study was conducted in various hospitals of Lahore. Informed consent was taken. The risk and benefits were explained and the personal information of study subjects was kept confidential. At 95% confidence level the sample size was calculated to be 100. The sample consisted of 50 cases and 50 controls. It was 1:1. Sampling was done by Purposive Method (non-probability). **Oral Clefts:** Oral cleft is a fissure in upper lip causing cleft lip and an opening in roof of mouth forming cleft palate.[18] **Syndromic Oral Clefts:** Oral clefts which are part of known congenital syndromes. **Non-syndromic Oral Clefts:** Oral clefts associated with no or one major anomaly or two or fewer minor anomalies.[19] **Case:** A patient of non-syndromic oral clefts having age of less than 5 years. **Control:**

Any child without any craniofacial anomaly having age of less than 5 years. **Cornell Medical Index (CMI):** It is a standardized questionnaire which is appropriate for collection of relevant medical and psychiatric information on a large mass with a less consumption of time of a doctor.[20] **Social Support Rating Scale:** A standardized questionnaire for collecting pertinent social support data.[21]

The children less or equal to 5years of age were divided in two groups. Patients having Cleft Lip, Cleft Palate or Cleft Lip and Palate is termed as a Case group while patients not having Cleft Lip, Cleft Palate or Cleft Lip and Palate constitute Control group. Patients having single minor associated anomalies except other craniofacial malformations (Patent Ductus Arteriosus, Inguinal hernia, Umbilical hernia, Undescended testes, Hydrocele).[22] The cases where consent was not granted by the parents are excluded from the study. Pretested and validated tools i.e. Cornell Medical Index Health Questionnaire and Social Support Performa were used. All data was collected , entered and analyzed with the help of SPSS version 20.0. Value of p less than 0.05 was considered as significant.

Results

56% of cases were females and 44% were males. Mean age of mothers was 27 and fathers was 31. Most mothers were uneducated (36%) while most fathers were educated till matric (34%). Income of most parents was in the range of 10,000-20,000 PKR (44%). 24% of controls were females and 76% were males.

The associations of oral cleft was found significant with parental respiratory, cardiovascular, gastrointestinal, musculoskeletal, skin-related, genitourinary health, easy fatigability, health habits, anxiety, anger, paternal nervous system health, sensitivity, and maternal frequency of illness, depression and feeling of inadequacy ($p < 0.05$). Maternal social support categories indicated that cases were less supported than controls: number of persons available and level of satisfaction ($p < 0.05$).

Table 1: Statistical Analysis of Cornell Medical Indices and Social Support Scores for Males.

Pair No.	System Studied	Paired Differences					t	df	Sig. (2-tailed)
		Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference				
					Lower	Upper			
1	Eyes and Ears (cases)- Eyes and Ears (controls)	.70 .60	.839 .904	.119 .128	-.255	.455	.566	49	.574
2	Respiratory(cases)- Respiratory (controls)	1.40 .64	1.429 1.102	.202 .156	.223	1.297	2.843	49	.007
3	Cardiovascular (cases)- Cardiovascular (controls)	1.54 .60	1.403 .833	.198 .118	.520	1.360	4.502	49	.000
4	Gastrointestinal (cases)- Gastrointestinal (controls)	1.56 .78	1.541 1.093	.218 .155	.272	1.288	3.085	49	.003
5	Musculoskeletal (cases)- Musculoskeletal (controls)	.82 .32	1.044 .741	.148 .105	.110	.890	2.573	49	.013
6	Skin (cases)-Skin (controls)	.66 .20	.872 .606	.123 .086	.160	.760	3.086	49	.003
7	Nervous (cases)- Nervous (controls)	1.04 .48	1.355 .614	.192 .087	.153	.967	2.767	49	.008
8	Genitourinary (cases)- Genitourinary (controls)	1.02 .38	1.220 .780	.173 .110	.239	1.041	3.208	49	.002

9	Fatigability (cases)- Fatigability (controls)	1.08 .36	1.291 .776	.183 .110	.250	1.190	3.078	49	.003
10	Frequency of illness (cases)- Frequency of illness (controls)	.52 .30	.863 .839	.122 .119	-.096	.536	1.399	49	.168
11	Miscellaneous diseases (cases)-Miscellaneous disease (controls)	1.04 .58	1.177 .835	.167 .118	.105	.815	2.605	49	.012
12	Health habits(cases)-Health habits (controls)	1.08 .58	1.322 .859	.187 .122	.054	.946	2.255	49	.029
13	Inadequacy (cases)- Inadequacy (controls)	.68 .64	1.058 1.083	.150 .153	-.341	.421	.211	49	.834
14	Depression(cases)- Depression (controls)	.46 .32	.734 1.096	.104 .155	-.214	.494	.795	49	.431
15	Anxiety (cases)- Anxiety (controls)	.74 .24	1.175 .687	.166 .097	.110	.890	2.573	49	.013
16	Sensitivity (cases)- Sensitivity (controls)	.86 .30	1.050 .839	.148 .119	.192	.928	3.055	49	.004
17	Anger (cases)- Anger (controls)	1.94 1.18	1.845 1.781	.261 .252	.137	1.383	2.453	49	.018
18	Tension (cases)- Tension (controls)	.70 .22	1.055 .616	.149 .087	.149	.811	2.914	49	.005

Table 2: Statistical Analysis of Cornell Medical Indices and Social Support Scores for Females.

Pair No.	System Studied	Paired Differences					t	df	Sig.(2-tailed)
		Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference				
					Lower	Upper			
1	Eyes and Ears (cases)- Eyes and Ears (controls)	1.04 .82	1.245 1.101	.176 .156	-.233	.673	.976	49	.334
2	Respiratory (cases)- Respiratory (controls)	1.28 .68	1.144 1.133	.162 .160	.127	1.073	2.547	49	.014
3	Cardiovascular (cases)- Cardiovascular (controls)	1.98 1.12	1.237 1.304	.175 .184	.401	1.319	3.762	49	.000
4	Gastrointestinal (cases)- Gastrointestinal (controls)	1.90 1.02	1.359 1.116	.192 .158	.346	1.414	3.309	49	.002
5	Musculoskeletal (cases)- Musculoskeletal (controls)	1.04 .60	1.261 .833	.178 .118	.025	.855	2.132	49	.038
6	Skin (cases)- Skin (controls)	.88 .30	1.206 .707	.171 .100	.154	1.006	2.735	49	.009
7	Nervous (cases)- Nervous (controls)	1.56 1.14	1.264 1.355	.179 .192	-.119	.959	1.566	49	.124

8	Genitourinary (cases)-Genitourinary (controls)	1.66 .88	1.437 1.172	.203 .166	.253	1.307	2.974	49	.005
9	Fatigability (cases)-Fatigability (controls)	1.30 .56	1.389 .837	.196 .118	.257	1.223	3.078	49	.003
10	Frequency of illness (cases)-Frequency of illness (controls)	.68 .20	1.203 .535	.170 .076	.130	.830	2.753	49	.008
11	Miscellaneous diseases (cases)-Miscellaneous disease (controls)	1.28 .92	1.230 .986	.174 .140	-.065	.785	1.703	49	.095
12	Health habits (cases)-Health habits (controls)	.96 .32	1.124 .653	.159 .092	.252	1.028	3.311	49	.002
13	Inadequacy (cases)-Inadequacy (controls)	1.48 .88	1.374 1.304	.194 .184	.061	1.139	2.239	49	.030
14	Depression (cases)-Depression (controls)	1.20 .48	1.325 .886	.187 .125	.233	1.207	2.969	49	.005
15	Anxiety (cases)-Anxiety (controls)	1.28 .60	1.371 1.107	.194 .156	.152	1.208	2.590	49	.013
16	Sensitivity (cases)-Sensitivity (controls)	1.48 .94	1.418 1.391	.200 .197	-.071	1.151	1.776	49	.082
17	Anger (cases)-Anger (controls)	1.40 .66	1.325 .982	.187 .139	.243	1.237	2.994	49	.004
18	Tension (cases)-Tension (controls)	1.26 .74	1.226 1.084	.173 .153	.056	.984	2.253	49	.029
19	Number of Persons available (cases)-Number of persons available (maximum:9) (controls)	1.1082 1.4552	.50769 .46933	.07180 .06637	-.53325	-.16075	-3.744	49	.000
20	Level of Satisfaction (cases)-Level of Satisfaction (maximum:6) (controls)	4.9318 5.4628	.79474 .40039	.11239 .05662	-.77732	-.28468	-4.332	49	.000

Discussion

Oral clefts have been investigated by scholars from various fields. Our research on Pakistani population shows that status of health of both parents and moral support of family and society plays a leading role in determination of oral clefts.

Maternal and paternal ages are believed to be independent risk factors for oral clefts. Existing literature proposes an association between increasing paternal age and the risk but maternal age has not been highlighted as such.[5-7] Our results also suggest an inclination along the same lines. However, we also find it mentionable that the data we collected has not been extensive enough to establish paternal age as a definite risk factor. Parental occupational exposures[16] and low socioeconomic status are also considered as risk factors for oral clefts[13]. However, such effects were not observed in our study.

Although many researchers have pointed out other risk factors like alcohol consumption, smoking, and specific vitamin deficiencies,[13,14] generalized parental health has rarely been discussed. In this study, it was seen that case parents had consistently poorer health status observed for each organ system subcategory. Studies conducted in past showed that smoking,[13] domestic air pollutants (e.g. stoves), commercial inha-

lants[16] may be perceived as a provoking factor in causing respiratory problems and our study also showed promising results. Problems of musculoskeletal, cardiovascular, skin – related and genitourinary health and illness frequency also may be considered as indicators for the generalized health of parents and are probably coupled with excessive physical work, approach to health care parental health and availability of medicines. Maternal stress has also been discussed as a risk factor for such congenital anomalies.[24] All of its possible causes can lead to raised blood glucocorticoid levels and high blood steroids may be linked to the risk of oral clefts.[23] Maternal stress levels were explored by assessing maternal perception of her own mental health and behavior as well as by employing the Social Support questionnaire. The results were found to be significant enough to consider the social support during pregnancy as a contributor towards the said risk.

As predictable for most case control studies, especially in case of less commonly encountered health problems, the main limitations of this study have been smaller sample size and recall bias. The smaller sample size reduces the ability to comment surely upon the debatable risk factors like parental age. The children with oral clefts require early surgical intervention mostly and hence present to the hospital at an earlier age. The same may not be true for the controls. Moreover, the parental educational status, understanding and memory regarding their health status during the pregnancy under discussion had to be relied upon and there remains the possibility of discrepancies in the accuracy on these accounts among the cases and controls. However, each step was carefully carried out to allow minimum hampering of accurate data collection. Certain measures were taken like explaining the questionnaires to the patients in their own language and according to their level of understanding and asking questions in more than one ways, to ensure that the exercise would be reliable enough.

Conclusion

Oral clefts have a multi-factorial etiology. Hence, an effective preventive program should take into account parental health and social support factors.

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