Medical Science

Authors' Affiliation:
[1]Preventive medicine resident doctor, Saudi board of preventive medicine, Jeddah, Saudi Arabia
[2]Consultant of preventive medicine and public health, Saudi board of preventive medicine, Ministry of health, Jeddah, Saudi Arabia

[*]Corresponding author
Preventive medicine resident doctor, Saudi board of preventive medicine,
Jeddah, Saudi Arabia
Email: Arwa.Alahmadi2@gmail.com

DISCOVERY

SCIENTIFIC SOCIETY

Knowledge and practice of primary healthcare physicians toward diagnosis of oral clefts through prenatal ultrasound imaging: An analytical cross-sectional study

Arwa Alahmadi[1*], **Majed Alghamdi**[2]

ABSTRACT

Background: Oral clefts are of the common malformation in newborns. The prevalence of this condition varies worldwide. Numerous negative consequences are linked to oral clefts and affecting both a child and parents. The influence can be minimized through early detection, and multidisciplinary team management. *Methods:* This is an analytical cross-sectional study that utilized an online questionnaire to assess primary healthcare (PHC) physician's knowledge about and practice toward prenatal diagnosis of oral clefts. *Results:* A total of 237 physicians with diversity in gender, age, and seniority level were included in the study. Overall knowledge and practice were insufficient in the sample. Several factors were significantly associated with level of knowledge and practice such as seniority level of the physician, and the hospital to which a physician reports (*p*-value<.05). *Conclusion:* Oral clefts are a devastating abnormality affecting children and parents life. An obvious paucity in knowledge and practice is observed among PHC physicians, and this necessitates a comprehensive training to establish competent PHC physicians in prenatal diagnosis of oral clefts.

Keywords: oral clefts, cleft lip, cleft palate, antenatal, primary healthcare

1. INTRODUCTION

Numerous malformations are recognized in newborns, and the most common craniofacial malformation in newborns is oral clefts. There are three main types of oral clefts: cleft lip alone, cleft lip with cleft palate, and cleft palate alone (Vyas et al., 2020). Oral clefts can present alone without any syndromic causes, which is often, or can be associated with other abnormalities. However, syndromic cases are associated with 30% cleft lip and palate conditions and 50% cleft palate malformations (Shi et al., 2008). The

prevalence of oral clefts differs between countries; the highest prevalence worldwide was identified in the First Nations (Amerindian) British Columbia, Canada (3 per 1000) (Lowry et al., 2009). Additionally, it differs based on race and ethnicity. For example, oral clefts are more common in Asians than Africans (Mai et al., 2014). The number of births in Saudi Arabia in 2020 was 585.17 thousand (Saudi Arabia Number of births, 2021). However, the prevalence of cleft lip and palate in Saudi Arabia is not well estimated in the literature, and there is a gap between studies in the matter of time of conducting the study. However, a systematic review discussed cleft lip and palate prevalence in Saudi Arabia and other Middle East countries included eight studies. The prevalence varied from 3 to 24 per 10000 live births (Sabbagh et al., 2012).

Parents of children with cleft lip and palate have difficulties building relationships with their newborns (Despars et al., 2011). Also, those parents complain of negative psychological impacts, such as confusion, stress, guilt, loss of control, and sometimes complain of depression (Bradbury and Hewison, 1994; Rey-Bellet and Hohlfeld, 2004). The effect on the parent-infant relationship may vary according to oral cleft types, as parents of cleft palate infants are more satisfied than parents of cleft lip and palate are (Despars et al., 2011). Early Surgical repair can improve the parent-infant relationship and decrease maternal stress significantly (Bradbury and Hewison, 1994; Rey-Bellet and Hohlfeld, 2004; Grollemund et al., 2020). According to the literature, parents with the prenatal diagnosis of cleft lip and palate are better at coping with their newborn status and preparing well for management through having a good time interval from diagnosis and first surgical intervention (Grollemund et al., 2020).

Cleft lip and palate can be diagnosed prenatally at 13 to 14 weeks of gestational age by trans-abdominal ultrasound (Vyas et al., 2020). Once cleft lip and palate are detected, parents need a multidisciplinary team to receive counseling about their coming baby and discuss management plans during and after delivery (Vyas et al., 2020). Also, this intervention is essential for the referral to genetic expertise to detect any other abnormalities and confirm if it is a syndromic or not through prenatal and postnatal genetic testing (Vyas et al., 2020). Referral to neonatologist to attend during delivery and perform precise planning for newborn management through discussing surgical repair and make clear perception regarding the advantages, disadvantages, timing, and financial costs of management options (Vyas et al., 2020). It is vital to educate the parents about their infant condition and how they may have issues with feeding due to sucking and swallowing disorders; moreover, it is crucial to inform the parents about the probability of hearing and speech problems. In addition, such a child is prone to respiratory and ear infections. However, these issues can be addressed by arranging postnatal referrals with specialized healthcare providers, such as surgeons, dentists, ENT specialists, speech therapists, psychologists, and social workers (Vyas et al., 2020).

The role of diagnosing oral clefts prenatally and referring a case to the appropriate services is crucial and it is, primarily, the role of practitioners in primary health care. Indeed, several studies revealed findings necessitating further education for primary healthcare practitioners and pediatricians regarding the diagnosis, referral and management of cleft lip and palate cases (Tanna et al., 2010; Lewis et al., 2017; Alharbi et al., 2019). Therefore, this study aims to assess the primary healthcare physician's knowledge about and practice toward prenatal diagnosis of oral clefts through ultrasound imaging in primary healthcare centers in Jeddah.

2. SUBJECTS AND METHODS

Study design and area
This is an analytical cross-sectional study that employed a web-based questionnaire for data collection. The study was carried out in Jeddah city in the Kingdom of Saudi Arabia. Data collection was accomplished through a web-based questionnaire targeting PHC physicians in Jeddah city, Saudi Arabia. There are, at the time of the study, 50 primary healthcare centers in Jeddah city, and subordinate to Ministry of Health general hospitals, which are King Abdullah medical city, King Fahad hospital, East Jeddah hospital, King Abdulaziz hospital, and Althagar hospital. Those primary healthcare centers serve all Jeddah population with diverse age groups, nations, and religions in an equitable manner. A primary healthcare center consists of several clinics that deliver primary servicessuch as antenatal clinics, well-baby clinics, dental clinics, family medicine clinics, and help people in the Bio-psycho-social model for prevention, referrals, and treatment of diseases.

Sample size and sampling technique
All physicians working at primary healthcare centers under MOH hospitals in Jeddah from March to June 2022 were eligible to be recruited in this study. Total number of physicians at PHC in Jeddah was 506 as stated in the 5-year statistics of MOH in 2018. The sample size was calculated using the Roasoft calculator and was set 241 after 10% added with a confidence level at 95% and a margin of error of 5%.

This study utilized a multi-stage random sampling technique. First, the investigators selected primary health care centers through stratified random sampling based on the five major MOH hospitals. Secondly, the investigators chose primary healthcare

centers from each stratum through simple random sampling. Finally, a simple random sampling was used to select potential participants from each included center.

Data collection procedure and tool

The data collection tool utilized in this study was a web-based questionnaire created in English language by the authors and it was face-validated by three experts in the field. The questionnaire included the following sections: demographic characteristics, basic knowledge information about how to diagnose cleft lip through antenatal ultrasound, and the third section contained five antenatal ultrasound images of different types of oral cleft cases to be assessed by the participants.

Ethical considerations

An ethical approval was obtained from the research ethical committee of the Research and Studies Department at the Directorate of Health Affairs in Jeddah city prior to data collection (No.H-02-J-002). A statement explaining the nature and the purpose of the study was included to gain the participants consent before filling the questionnaire. All data was handled anonymously and used for research purposes only.

Statistical analysis

Data analysis was carried out using IBM SPSS Statistics for MacOS, version 27 (IBM Corp., Armonk, N.Y., USA). All categorical variables were summarized as frequencies and percentages. Skewed distribution numerical variables were presented as median and interquartile range values. For bivariate analyses, Chi-Square test and the extension of Fisher-Freeman-Halton Exact test were used to analyze categorical data. A p-value of <0.05 was considered statistically significant.

3. RESULTS

A total of 237 physicians participated in the study and all of them were included in the analysis. More than 46% of the participating physicians fall in the 20 – 29 years old age category, and most participants (64.6%) were females. Eighty-nine (37.6%) individuals were working as a general practitioner, and most participants have a working experience of ≤5 years (Table 1).

Table 1 Participant Demographic Characteristics

n=237		n	%
Gender	Male	84	35.4%
	Female	153	64.6%
Age	20 – 29	110	46.4%
	30 – 39	86	36.3%
	40 – 49	34	14.3%
	50 – 60	7	3.0%
Position	General practitioner (GP)	89	37.6%
	Resident	78	32.9%
	Family medicine specialist	47	19.8%
	Family medicine consultant	23	9.7%
Experience (years)	5 or less	139	58.6%
	6 – 9	52	21.9%
	10 or more	46	19.4%
Hospital name	King Abdullah medical city	80	33.8%
	East Jeddah hospital	46	19.4%
	Althagar hospital	34	14.3%
	King Fahad hospital	40	16.9%
	King Abdulaziz hospital	37	15.6%

The study asked participants to rank their confidence in interpreting ultrasound images of oral cleft cases from "1" (lowest) to "5" (highest). Apparently, 71% of the physicians included in the study ranked their confidence as "1" indicating lowest level of

confidence (Figure 1). Nevertheless, the study participants reported their perception of usefulness of oral cleft diagnosis during antenatal care visits in primary healthcare centers, and the higher proportion (34.2%) perceived antenatal diagnosis of oral clefts as very useful (Figure 2).
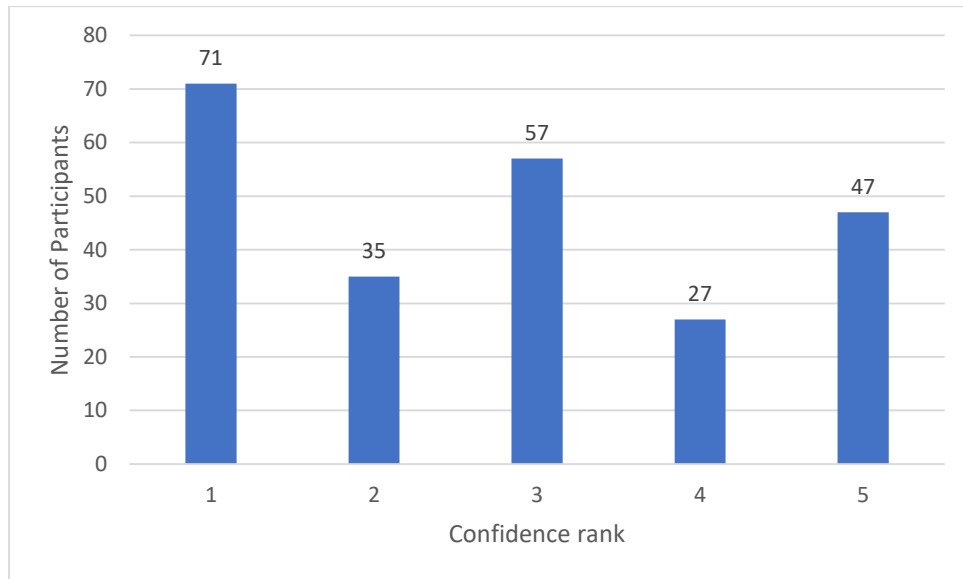

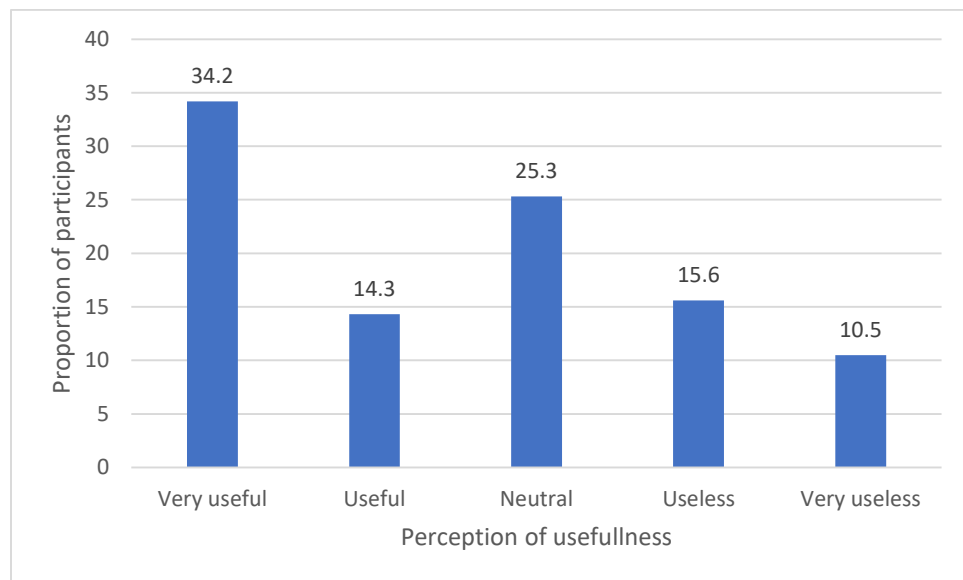**Figure 1** Participants confidence in diagnosing oral clefts in prenatal care

Figure 2 Participants perception of oral clefts diagnosis usefulness

The participant's knowledge was assessed through multiple questions and the achievable score ranged from 0 to 19 points. The median knowledge score was 10 with an interquartile range of 6 points. The knowledge score was categorized into insufficient and sufficient, if the participant achieved 60% or more of the correct answers, they were considered to show sufficient level of knowledge. Table 3 illustrates the associated factors with knowledge level of the participating physicians. Only the working position and place of work showed statistically significant association with the knowledge level (p=.001, .017 respectively). Other factors seem to have an association that is not statistically significant.

Table 3 Factors Associated with Knowledge Level

n=237		Knowledge Level				
		Insufficient		Sufficient		
		n	%	n	%	*p*-value
Gender	Male	47	56.0%	37	44.0%	.68
	Female	81	52.9%	72	47.1%	
Age	20 – 29	59	53.6%	51	46.4%	.83
	30 – 39	49	57.0%	37	43.0%	
	40 – 49	17	50.0%	17	50.0%	
	50 – 60	3	42.9%	4	57.1%	
Position	General practitioner (GP)	59	66.3%	30	33.7%	.001*
	Resident	42	53.8%	36	46.2%	
	Family medicine specialist	22	46.8%	25	53.2%	
	Family medicine consultant	5	21.7%	18	78.3%	
Experience (years)	5 or less	74	53.2%	65	46.8%	.38
	6 – 9	32	61.5%	20	38.5%	
	10 or more	22	47.8%	24	52.2%	
Hospital	King Abdullah medical city	32	40.0%	48	60.0%	.017*
	East Jeddah hospital	30	65.2%	16	34.8%	
	Althagar hospital	20	58.8%	14	41.2%	
	King Fahad hospital	27	67.5%	13	32.5%	
	King Abdulaziz hospital	19	51.4%	18	48.6%	
Chi-Square test, Fisher-Freeman-Halton Exact test						
*Significant						

Several case scenarios of oral cleft were presented to the participants to assess their practice and the achievable score ranged from 0 to 20 points. The median practice score was 6 with an interquartile range of 5 points. For better understanding, the practice score was categorized into insufficient and sufficient, if the participant achieved 60% or more of the correct answers, they were considered to show sufficient level of practice. All independent factors were studied for association with the level of practice, however only the organization to which a participant reports showed statistically significant association (*p* =.029) (table 4).

Table 4 Factors Associated with Practice Level

n=237		Practice Level				
		Insufficient		Sufficient		
		n	%	n	%	*p*-value
Gender	Male	75	89.3%	9	10.7%	.2
	Female	144	94.1%	9	5.9%	
Age	20 – 29	102	92.7%	8	7.3%	.81
	30 – 39	80	93.0%	6	7.0%	
	40 – 49	30	88.2%	4	11.8%	
	50 – 60	7	100.0%	0	0.0%	
Position	General practitioner (GP)	85	95.5%	4	4.5%	.15
	Resident	73	93.6%	5	6.4%	
	Family medicine specialist	42	89.4%	5	10.6%	
	Family medicine consultant	19	82.6%	4	17.4%	
Experience (years)	5 or less	130	93.5%	9	6.5%	.34
	6 – 9	49	94.2%	3	5.8%	
	10 or more	40	87.0%	6	13.0%	
Hospital name	King Abdullah medical city	68	85.0%	12	15.0%	.029*

	East Jeddah hospital	43	93.5%	3	6.5%	
	Althagar hospital	33	97.1%	1	2.9%	
	King Fahad hospital	40	100.0%	0	0.0%	
	King Abdulaziz hospital	35	94.6%	2	5.4%	
Chi-Square test, Fisher-Freeman-Halton Exact test						
*Significant						

4. DISCUSSION

This study aims to assess the knowledge regarding and practice toward oral cleft lip and palate among primary healthcare (PHC) physicians in Jeddah. The study findings show that more than 50% of PHC physicians have insufficient knowledge regarding oral cleft lip and plate as 128 of 237 participants (54%) scored below 60% of the assessment tool used in the current study. Certain factors showed statistically significant association with the level of knowledge. One of these factors is the physician seniority or educational level as most family medicine consultants (78.3%) working in the PHC showed a sufficient level of knowledge during the assessment. Similarly, 53.2% of family medicine specialists scored more than 60% in the knowledge assessment tool. On the other hand, less than 50% of the training residents and general practitioners in the PHC achieved a sufficient level of knowledge during the study assessment. This may indicate how education and training influence the physician knowledge, thus how they manage their patients. Moreover, the proportion of physicians working in PHCs reporting to King Abdullah medical city seem to achieve sufficient knowledge more than their peers in other PHCs, however this variation may be attributable to the fact that King Abdullah medical city physicians account for most participants in the current study, and this may be influenced by the fact that the number of training residents was higher at King Abdullah medical city than it was in other centers. Although 60% of participants from King Abdullah medical city showed sufficient knowledge regarding prenatal diagnosis of oral cleft lips and palates, the practice score for the same cohort was insufficient in 85% of them. In fact, most participants in the study failed to achievea score of 60% or more in the practice assessment, and this overall image might be explained by several reasons such as unfamiliarity with such cases, ambiguity as ultrasound images were shown on electronic devices screens, or the difficulty level of the assessment tool.

The findings of the current study show a universal lack in knowledge and practice of PHC physicians, and this paucity may result in an incompetence and improper therapeutic management, subsequently. Indeed, among the included physicians the vast majority rate their confidence in diagnosing oral clefts "lowest" regardless of the physician seniority. Although a thorough search in the literature was carried out by the study investigators, the search failed to discover articles assessing the knowledge of the PHC practitioners. However, a large survey study was conducted in 2018 in which a total of 1435 primary care physicians were included, the study investigated the experience, knowledge, and confidence of the included sample in managing and dealing with oral cleft children. The studies showed that most physicians had limited training and experience with oral cleft patients and were more confident in providing routine care to these patients, yet not very comfortable in providing counseling to the parents regarding oral cleft-related concerns.

Furthermore, the study participants showed a preference and comfort in managing oral cleft children with the availability of multidisciplinary expertise (Damiano et al., 2010). When using ultrasound for screening of cleft lips and palates, ultrasonography performers are required to have a comprehensive understanding of face embryology to enhance the effectiveness of the prenatal screening (Smarius et al., 2017). Having good knowledge of the embryology of the face helps practitioners work efficiently and make an accurate test. In fact, several studies shed light on the need for further education and training of primary healthcare practitioners to enhance their ability in diagnosis, treatment and referral of oral cleft patients (Tanna et al., 2010; Lewis et al., 2017; Alharbi et al., 2019). The primary care physician knowledge and experience in oral cleft management is crucial. A study including 235 oral cleft children compared the perception and outcomes between parents who received diagnosis prenatally and parents who received it at birth. Interestingly, the study results showed no significant difference between the two groups in terms of satisfaction and outcomes.

Nevertheless, mothers whose children were diagnosed prenatally were more likely to rate the healthcare providers more moderately or very helpful in facilitating infant feeding process than mothers who knew the diagnosis at birth. In addition, most participating parents reported they wished for more comprehensive and rich information regarding the infant condition, obstacles and healthcare services available for such children (Robbins et al., 2010). Similarly, another study agrees with these findings as mothers stated they were in need of much knowledge and information about oral clefts in addition to the appropriate emotional support and referral at the time of diagnosis (Hlongwa and Rispel, 2018). Such incomprehensive knowledge may lead to lack of proper management such as lack of referral to appropriate team and specialties.

A study compared the outcomes of oral cleft patients between those who received team care and those who received individual provider care. The aforementioned study findings show that patients without team care were less likely to receive genetics counseling, hearing tests, and dental care visits (Austin et al., 2010). It is obvious how much risk an oral cleft patient may face through their lifetime, thus a lack of referral and such tiny mistakes must not add to the existing challenges.

Recommendations

The burden of oral cleft conditions is huge on patients, families, and healthcare providers. As primary health care is the cornerstone for the healthcare system, and it is the first encounter where an oral cleft case can be diagnosed, it is vital for the primary healthcare physicians to gain the required competencies to identify such cases. In fact, the PHC practitioners may influence the management substantially through coordinating the healthcare services for oral cleft patient's viareferral to specialized healthcare services and make the best use of the multidisciplinary team management. The authors noticed through their experience in PHC centers shortagein practitioners who are trained on prenatal ultrasonography, thus they recommend expanding the training to overcome this paucity and have at least one trained physician who is competent to diagnose oral cleft cases prenatally.

5. CONCLUSION

This study included 237 primary healthcare physicians to assess their knowledge about and practice toward prenatal diagnosis of oral cleft lips and palates. The participated physicians were of both genders and from various seniority levels as it included general practitioners, residents, specialists, and consultants. The findings show a global insufficiency in both knowledge and practice of the participants. Therefore, a professional and comprehensive training is advised for primary healthcare practitioners in order to establish enough competent practitioners in primary healthcare centers, hence improving the service provided for such patients and ultimately promoting their prognosis.

Abbreviations

3D-US: Three-dimensional ultrasound
2D-US: Two-dimensional ultrasound
GERD: Gastroesophageal reflux disease
MOH: Ministry of health
PHC: primary healthcare centers
ENT: ear, nose and trachea
SPSS: statistical package for social science

Author's contributions

Author AA established the literature search, managed the data collection and analysis, and prepared the first draft of the manuscript. Author MA participated intellectually in the research and reviewed the final draft of the manuscript.

Ethical approval

An ethical approval was obtained from the research ethical committee of the Research and Studies Department at the Directorate of Health Affairs in Jeddah city prior to data collection (Ethical approval code: H-02-J-002).

Conflicts of interest

The authors declare that there are no conflicts of interests.

Data and materials availability

All data associated with this study are present in the paper.

REFERENCES AND NOTES

1. Alharbi AA, Al-Thunayyan FS, Alsuhaibani KA, Alharbi KA, Alharbi MA, Arkoubi AY. Perception of primary health care providers of plastic surgery and its influence on referral. J Fam Med Prim care 2019; 8(1):225–230. doi: 10.4103/jfmpc.jfmpc_204_18

2. Austin AA, Druschel CM, Tyler MC, Romitti PA, West II, Damiano PC, Robbins JM, Burnett W. Interdisciplinary craniofacial teams compared with individual providers: is orofacial cleft care more comprehensive and do parents perceive better outcomes? Cleft Palate-Craniofacial J 2010; 47(1):1–8 doi: 10.1597/08-250.1

3. Bradbury ET, Hewison J. Early parental adjustment to visible congenital disfigurement. Child Care Health Dev 1994; 20(4):251–266 doi: 10.1111/j.1365-2214.1994.tb00388.x

4. Damiano PC, Tyler MC, Romitti PA, Druschel C, Austin AA, Burnett W, Robbins JM. Primary care physician experience with children with oral clefts in three states. Birth Defects Res Part A Clin Mol Teratol 2010; 88(12):1050–1056 doi: 10.1002/bdra.20726

5. Despars J, Peter C, Borghini A, Pierrehumbert B, Habersaat S, Müller-Nix C, Ansermet F, Hohlfeld J. Impact of a cleft lip and/or palate on maternal stress and attachment representations. Cleft Palate-Craniofacial J 2011; 48(4):419–424 doi: 10.1597/08-190

6. Grollemund B, Dissaux C, Gavelle P, Martínez CP, Mullaert J, Alfaiate T, Guedeney A. The impact of having a baby with cleft lip and palate on parents and on parent-baby relationship: the first French prospective multicentre study. BMC Pediatr 2020; 20(1):230 doi: 10.1186/s12887-020-02118-5

7. Hlongwa P, Rispel LC. People look and ask lots of questions: Caregivers' perceptions of healthcare provision and support for children born with cleft lip and palate. BMC Public Health 2018; 18(1):1–10 doi: 10.1186/s12889-018-5421-x

8. Knoema. Saudi Arabia Number of births, 1950-2020. 2021. Available from: https://knoema.com/atlas/Saudi-Arabia/topics/Demographics/Fertility/Number-of-births?mode=amp

9. Lewis CW, Jacob LS, Lehmann CU. The primary care pediatrician and the care of children with cleft lip and/or cleft palate. Pediatrics 2017; 139(5) doi: 10.1542/peds.2017-0628

10. Lowry RB, Johnson CY, Gagnon F, Little J. Segregation analysis of cleft lip with or without cleft palate in the First Nations (Amerindian) people of British Columbia and review of isolated cleft palate etiologies. Birth Defects Res A Clin Mol Teratol 2009; 85(6):568–73 doi: 10.1002/bdra.20558

11. Mai CT, Cassell CH, Meyer RE, Isenburg J, Canfield MA, Rickard R, Olney RS, Stallings EB, Beck M, Hashmi SS, Cho SJ, Kirby RS. Birth defects data from population-based birth defects surveillance programs in the United States, 2007 to 2011: highlighting orofacial clefts. Birth Defects Res Part A Clin Mol Teratol 2014; 100(11):895–904 doi: 10.1002/bdra.23329

12. Rey-Bellet C, Hohlfeld J. Prenatal diagnosis of facial clefts: evaluation of a specialised counselling. Swiss Med Wkly 2004; 134(43–44):640–4 doi: 2004/43/smw-10547

13. Robbins JM, Damiano P, Druschel CM, Hobbs CA, Romitti PA, Austin AA, Tyler M, Reading JA, Burnett W. Prenatal diagnosis of orofacial clefts: association with maternal satisfaction, team care, and treatment outcomes. Cleft Palate-Craniofacial J 2010; 47(5):476–481 doi: 10.1597/08-177

14. Sabbagh HJ, Mossey PA, Innes NPT. Prevalence of orofacial clefts in Saudi Arabia and neighboring countries: a systematic review. Saudi Dent J 2012; 24(1):3–10 doi: 10.1016/j.sdentj.2011.11.001

15. Shi M, Wehby GL, Murray JC. Review on genetic variants and maternal smoking in the etiology of oral clefts and other birth defects. Birth Defects Res C Embryo Today 2008; 84(1):16–29 doi: 10.1002/bdrc.20117

16. Smarius B, Loozen C, Manten W, Bekker M, Pistorius L, Breugem C. Accurate diagnosis of prenatal cleft lip/palate by understanding the embryology. World J Methodol 2017; 7(3):93–100 doi: 10.5662/wjm.v7.i3.93

17. Tanna N, Patel NJ, Azhar H, Granzow JW. Professional perceptions of plastic and reconstructive surgery: what primary care physicians think. Plast Reconstr Surg 2010; 126(2):643–650 doi: 10.1097/PRS.0b013e3181de1a16

18. Vyas T, Gupta P, Kumar S, Gupta R, Gupta T, Singh H. Cleft of lip and palate: a review. J Fam Med Prim Care 2020; 9(6):2621 doi: 10.4103/jfmpc.jfmpc_472_20