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## REVIEW ARTICLE

# Prevalence of orofacial clefts in Saudi Arabia and neighboring countries: A systematic review

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### KEYWORDS

Prevalence;  
Saudi Arabia;  
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Middle East

**Abstract** Cleft lip and/or palate are among the most common birth defects in the world. The prevalence of these conditions varies considerably across geographic areas and ethnic groups.

**Objective:** The aim of this study was to carry out a systematic review and appraisal of the literature on the prevalence of cleft lip and/or palate in Saudi Arabia and comparable Middle Eastern countries.

**Materials and methods:** All published articles on orofacial clefts (OFC) in Saudi Arabia and its bordering countries in the Middle East with similar and comparable population characteristics were reviewed in July 2010.

**Results:** After reviewing the articles, only eight matched the inclusion criteria. Three studies were carried out in two regions in Saudi Arabia (Riyadh and Al-Qaseem). The other five studies were set in Dubai, Oman, and Jordan. The prevalence of cleft lip and/or palate reported in these studies varied greatly from 0.3 to 2.4 per 1000 live births. The birth prevalence of orofacial clefts in males was reported to be higher than in females. The isolated cleft palate prevalence was reported to be higher in females in most of the studies.

**Conclusion:** The eightfold variation in the prevalence of orofacial clefts between highest and lowest prevalence is likely to be due, at least in part, to problems with ascertainment, but there may also be underlying genetic or environmental factors that require further investigation.

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## 1. Introduction

Orofacial clefting (OFC) describes a spectrum of disorders from partial or complete fissuring of the upper lip, with or without fissuring of the palate [i.e., cleft lip and palate (CLP) or cleft lip (CL)] to fissuring of the palate alone [i.e., isolated cleft palate (CP)] (Mossey and Castilla, 2001). These features may present alone, as part of a syndrome, or along with other associated abnormalities (Mossey et al., 2009). Collectively, OFCs are known to be the most common craniofacial defects and one of the most common structural birth defects throughout the world (Christensen et al., 2004). The estimated overall global birth prevalence of OFC is one affected individual in every 700 newborn babies (World Health Organization, 2003). However, in spite of OFCs occurring in all races, the prevalence of individual OFC conditions vary considerably across geographic areas and ethnic groupings. For example, OFC more commonly occurs among Asian than African populations. It is important to understand the prevalence of craniofacial anomalies in every community to determine the size of the problem, the effort needed to improve the quality of life of these patients, and the efficacy of interventions. Even though efforts have been made to record the frequency of birth defects over the years, accurate epidemiological data do not exist for many countries (Mossey and Little, 2002).

In Saudi Arabia, nearly 355,000 children are born each year (Ministry of Health, 2008). There is a high rate of consanguineous marriage, which could be indicative of a high prevalence of congenital anomalies (Narchi and Kulaylat, 1997). However, there is currently no national register for OFC prevalence in Saudi Arabia. Therefore, a systematic review of the literature for the Saudi population was undertaken to search for information on the prevalence of OFC, which might give a clearer picture. The literature search was expanded to include studies carried out in other Middle Eastern countries with similar and comparable population characteristics to allow for comparisons and to provide context. Therefore, the aim of this

study was to carry out a systematic review and appraisal of the literature on the prevalence of OFC in Saudi Arabia and comparable Middle Eastern countries.

## 2. Materials and methods

### 2.1. Protocol for the literature search

A protocol for the systematic assessment of the literature on the prevalence of OFC in Saudi Arabia and other Middle Eastern countries was developed. The Middle Eastern countries included were: Jordan, Syria, UAE, Qatar, Kuwait, Bahrain, Oman, and Yemen. All of these countries border Saudi Arabia. The populations of these countries are comparable to the Saudi population in terms of the following characteristics: ethnic group, religion, tradition, culture, high rate of consanguineous marriage, and high birth rate. Iraq was excluded because of the suspected high prevalence of teratogenic defects resulting from long-term conflict (Fathallah, 2007).

The literature search protocol that was formulated consisted of the following: keyword identification, development of a search strategy, selection of search engines, and definition of inclusion/exclusion criteria for identified studies. The search strategy consisted of combinations of three keyword groups: (1) prevalence, epidemiology; (2) orofacial cleft, cleft palate, cleft lip, craniofacial anomalies; and (3) Jeddah, Riyadh, Saudi Arabia, Middle East, Arabia, Jordan, Syria, United Arab Emirates, UAE, Qatar, Kuwait, Bahrain, Oman, and Yemen. The search engines used were PubMed and Scopus. In addition, certain keywords were used to search Google (from the year 1980 to 2010), the Saudi Dental Journal (SDJ), the Saudi Medical Journal (SMJ), Jordan Medical Journal, Syrian American Medical Journal, Avicenna Journal of Medicine, Syria Medical Journal, Kuwait Medical Journal, International Arab Journal, Oman Medical Journal, Yemeni Journal for Medical Sciences, Bahrain Medical Bulletin, Journal of Bahrain Medical Society, and Qatar Medical Journal. The searches

were performed in July 2010. All papers were obtained, websites were checked, and references for articles and websites were reviewed for other relevant articles and abstracts. The search did not exclude any languages. In cases where data were reported in more than one article, the data were only extracted once.

### 2.2. Inclusion and exclusion criteria for studies

All papers were reviewed for studies meeting the following inclusion criteria:

- The study reported data on the prevalence of cleft lip and palate in Saudi Arabia or one of the countries in the Middle East listed above.
- If the prevalence of cleft lip and palate was not included in the paper, then sufficient information to calculate an approximate prevalence of OFC was required. For instance, the timing and methods used for data collection and the sample size were required to allow for an estimation of the denominator size of the base population.

The exclusion criteria for studies were the following:

- The prevalence of OFC was not reported and could not be estimated from the presented data.
- The article reported information on the prevalence of craniofacial anomalies as a whole, but did not specify the prevalence of OFC.
- The study did not present sufficient details on the materials and methods.

### 2.3. Data extraction

Papers meeting the inclusion criteria were reviewed, and when available the following data were extracted independently and in duplicate by two of the authors (HS and NI):

- dates, geographic location, and design of the study;
- sample size, source of population, and nationalities;
- demographic data of the sample;
- prevalence of cleft lip and/or palate; and
- prevalence of cleft lip and/or palate according to sex, consanguineous marriage rate, the presence of positive family history, and the association of other deformities.

The prevalence of OFC from each study was recorded, and the mean was compared with global prevalence figures.

## 3. Results

### 3.1. Search strategy results

The search strategy produced the following results: 45 articles in PubMed, 13 articles in Scopus, 9 articles in the SDJ, and 3 articles in the SMJ. Google registered 97 hits. All articles were in English with most having an Arabic abstract.

After checking the references and excluding duplicated articles, there were 126 papers. The majority of the studies did not meet the inclusion criteria and were excluded because they only

described the characteristics and types of CL, CP, or CLP or discussed the prevalence of associated anomalies or diseases.

### 3.2. Studies fitting the inclusion criteria

Only eight articles fulfilled the inclusion criteria. Three studies were carried out in Saudi Arabia in two regions; two articles (Al-Johar et al., 2008; Kumar et al., 1991) and one report (Al-Johar et al., 2009) were performed in Riyadh, and one article (Borkar, 1993) was performed in Al-Qaseem. The remaining five were carried out in the following Middle Eastern countries: UAE (Al-Talabani et al., 1998), Oman (Patel, 2007; Rajab and Thomas, 2001), and Jordan (Al-Omari and Al-Omari, 2004; Aqrabawi, 2008). One study (Al-Johar et al., 2008) and one report (Al-Johar et al., 2009) covered the same location and time span. These were set in the King Faisal Specialised Hospital and Research Centre (KFSHRC), which is a well established cleft center in Riyadh. Data from these studies were combined to provide a figure for prevalence. This figure was determined by taking the reported data for OFC in the year 2008 (Al-Johar et al., 2009) and comparing it with the total number of reported live births (almost 80,000 live births per year) in the same year in Riyadh (Ministry of Health, 2008).

### 3.3. Prevalence of OFC

The prevalence figures for each study are shown in Table 1. The lowest prevalence of cleft lip and/or palate was 0.3 births per 1000 live births (Kumar et al., 1991; Al-Johar et al., 2009). Both studies with the lowest prevalence were carried out in Riyadh, Saudi Arabia. The highest prevalence was 2.4 per 1000 live births (Aqrabawi, 2008) in Jordan. The overall mean prevalence of OFC for all the studies was 1.25 per 1000 live births, which is close to the reported global prevalence of one in every 700 births (World Health Organization, 2003).

For the studies carried out in Saudi Arabia, the reported prevalence rates varied by almost a factor of 10, from 0.3 (Al-Johar et al., 2008, 2009) to 2.19 per 1000 live births (Borkar, 1993). Different strength and limitation points affected each study's ascertainment (see Table 3).

### 3.4. Characteristics of OFC

All studies reported a higher prevalence of CLP than other types of clefts (Table 1). In children with unilateral clefts, the left-sided defects were more common than right-sided defects. The prevalence of OFC in males was reported to be higher than in females with ratios for all clefts ranging from 1.2:1 to 4:1 and the ratios of CLP ranging from 1:1 to 1.8:1. On the other hand, isolated CP incidence was reported to be higher in females with a ratio of 3:1 except for one study (Aqrabawi, 2008) in which all the CP cases reported were males.

### 3.5. Associated anomalies

Six out of eight studies reported associated anomalies (Table 2). These associated anomalies had rates ranging from 12% (Borkar, 1993) to 58.4% (Rajab and Thomas, 2001). The most common associated anomaly reported was congenital heart disease (CHD), which was reported at rates of 29% (Borkar, 1993), 20% (Kumar et al., 1991), and 38.2% (Al-Johar et al.,

**Table 1** Included studies, their characteristics and prevalence of OFC.

Reference	Site and country	Duration	Study design	Population (Dominator)	Sample size	Population characteristics	Male%	Prevalence	CLP%	CL%	CP%
Kumar et al. (1991)	KKUH <sup>a</sup> , Riyadh, Saudi Arabia	1982–1988	Retrospective	KKUH: 20,045 live births/6 years	6 newborn/6 years	Mainly Saudis	–	0.3/1000 live births	–	–	–
Al-Johar et al. (2008) Al-Johar et al. (2009)	KFSHRC <sup>b</sup> , Riyadh, Saudi Arabia	1999–2009	Retrospective	Saudi Arabia: 300,000 births/year Riyadh: 80,000 births/year Al-Qasseem: 62,557 live births/4 years (MOH birth registry statistics)	1319/11year86/year from SA19/year from Riyadh	95% Saudis	56	0.3/1000 live births	47.8	15.7	36.5
Borkar (1993)	KFSH <sup>c</sup> , Al-Qasseem, Saudi Arabia	1989–1992	1 year retrospective + 3 year prospective	Al-Qasseem: 62,557 live births/4 years (MOH birth registry statistics)	137	All Saudis	–	2.19/1000 live births	45	41	14
Al-Talabani et al. (1998)	Corniche Hospital Abu Dhabi, UAE	1992–1995	Prospective	Corniche Hospital: 24,233 live and still births/3 years	13	73% Arabs	–	0.5/1000 live and still births	–	–	–
Rajab and Thomas (2001)	Khoula Hospital, and all maternity sections in Oman	1989–1995	Retrospective	Oman: 375,000 births in all 2ry and 3ry level hospitals/7 years (80% of total population)	563	All Omanis	51	1.5/1000 live births	41	23	36
Patel (2007)	10 health institution with maternity facilities and includes two 2ry care hospital Dhahire, Oman	2003–2005	Retrospective	10,311	14	93% Omanis	–	1.5/1000 live and still births	35	21	43
Al-Omari and Al-Omari (2004)	KHMC <sup>d</sup> and Al-Bashir Hospital, Amman, Jordan	1991–2001	Retrospective	Jordan: 1548,106 births/11 years	2146	All Jordanians	54	1.39/1000 live births	48	30	22
Aqrabawi (2008)	KHMC <sup>d</sup> Amman, Jordan	Jan 2000-Jan 2005	Prospective	25,440 live births/5 years	60	All Jordanians	75	2.4/1000 live births	45	20	15

<sup>a</sup> King Khalid University Hospital.

<sup>b</sup> King Faisal Specialised Hospital and Research Centre.

<sup>c</sup> King Fahad Specialised Hospital.

<sup>d</sup> King Hussein Royal Medical Centre.

**Table 2** Associated anomalies, consanguinity and positive family history for OFC, for included studies, where reported.

Study	Associated anomalies (%)	% of Consanguinity +ve Family history for OFC		1st degree cousin
		All relatives		
Kumar et al. (1991)	13.4	6.7	6	26.8%
Al-Johar et al. (2008)Al-Johar et al. (2009)	29.5	54.4	53.3	28%
Borkar (1993)	12	42	–	–
Rajab and Thomas (2001)	58.4	–	45	23%
Al-Omari and Al-Omari (2004)	18	–	–	–
Aqrabawi (2008)	47	83	–	None

2008). The Jordanian study (Aqrabawi, 2008) reported that 47% of all cleft patients had CHD.

### 3.6. Consanguinity

There were large differences in the reported consanguinity rates for parents of children with OFCs (Table 2). Five of the studies reported on sanguineous relationships. Four of them reported on consanguineous relationships with rates ranging from 6.7% (Kumar et al., 1991) to 83% (Aqrabawi, 2008). Marriages between first-degree cousins were reported in three studies in more than 80% of consanguineous cases with OFC (Kumar et al., 1991).

### 3.7. Positive family history of OFC

The frequency of family history of OFC was reported in four studies, with rates of 23% (Rajab and Thomas, 2001), 26.8% (Kumar et al., 1991), 28% (Al-Johar et al., 2008), and 0% (Aqrabawi, 2008).

## 4. Discussion

Understanding the epidemiology of birth defects can provide a basis for further investigation into the etiology and pathogenesis of these developmental disorders. However, complete ascertainment of the prevalence of affected individuals is difficult, and it is influenced by a number of factors. Among the main factors that affect ascertainment are the following: whether sampling is population or hospital based, the type and number of information resources available, the timing of data collection (immediately after birth or after several weeks), and whether stillbirths are included in addition to live births (Mossey and Castilla, 2001).

### 4.1. OFCs ascertainment: (see Table 3)

From this analysis of the literature on OFC in Saudi Arabia and its bordering countries, studies were only found for four countries (Oman, Jordan, UAE, and Saudi Arabia). In total, eight articles were published on the subject, of which three were from Saudi Arabia. The studies carried out in Saudi Arabia (Al-Johar et al., 2008; Borkar, 1993; Kumar et al., 1991) were confined to two cities (Riyadh and Al-Qaseem), with data being collected from one type of information source (hospital records). These studies did not include stillbirths. The records were from referral centers, not maternity hospitals, and the sites were not the only referral centers in these regions. There-

fore, these data were likely under-ascertained, and this is the most probable explanation for the differences in the reported prevalence of OFC in Saudi Arabia, which varies by ten-fold (0.3–2.19 per 1000 live births). It is not possible to say to what extent variability in methodologies used for data collection or differences in environmental factors between these two cities contribute to the reported figures. Therefore, based on these studies the prevalence of OFC cannot be clearly determined.

An exemplary project initiating registration of OFC anomalies in Saudi Arabia was carried out in KFSHRC, Riyadh, Saudi Arabia. In this project, 1319 patients with OFC and other craniofacial anomalies were registered between 1999 and 2009. The prevalence calculated from this study was 0.3 clefts per 1000 live births. However, this figure may be an underestimate of the true prevalence, as KFSHRC is not the only tertiary center in Riyadh for cleft lip and palate management (Al-Johar et al., 2008, 2009). Similarly, this could account for the low prevalence in the other Riyadh study (Kumar et al., 1991).

In the study in Al-Qaseem (Borkar, 1993), there was a high prevalence of OFC (2.19/1000). This prevalence was calculated by comparing children born with OFC and referred to King Fahad Specialised Hospital (KFSH) with the total population of children born in the same period and in the same region using Ministry of Health (MOH) statistical records. The authors claimed that their study represented the population of Al-Qaseem because KFSH was the only tertiary center in this area, and all five peripheral hospitals followed a rigid control system of referral. There are a number of possible reasons for the high prevalence figure. There may have been an underestimation in the denominator population figure because KFSH is a referral center with no maternity hospital. There was no clear explanation of the methodology involving the subjects' age, so it is possible that older children were included in the sample. The multiple conflicts in neighboring regions (US Department of Defense, 1998) may have had teratogenic effects. Additionally, there was a high percentage of families from rural areas, which has been reported to increase the risk of OFC (Al-Sahafi, 2010). Accordingly, the validity of the reported prevalence is unclear.

Three studies, two from Oman (Patel, 2007; Rajab and Thomas, 2001) and one from Jordan (Al-Omari and Al-Omari, 2004), reported a prevalence of OFC consistent with the WHO global report of one in every 700 births (World Health Organization, 2003). Both Rajab and Thomas (2001) and Al-Omari and Al-Omari (2004) had a large sample compared to the other studies, with a long period of data collection, and these studies were carried out at the only referral centers in their countries. The ascertainment and calculation of denominator data should



**Table 3** Methodological characteristics of included studies.

	Strengths	Limitations
Kumar et al., 1991	<ul style="list-style-type: none"> <li>Data collected from patients seen in a University hospital. Therefore, stands by itself with a fair representation of the population</li> </ul>	<ul style="list-style-type: none"> <li>Hospital based</li> <li>Retrospective study</li> <li>It is a university hospital, not a maternity hospital nor the only referral centre in Riyadh that provides OFC treatment</li> <li>Cannot be generalized to the population,</li> <li>Data obtained from birth records (one source)</li> <li>Midline cleft was included in the study</li> <li>Small sample size</li> <li>Prevalence in living births only</li> </ul>
Al-Johar et al., 2008 Al-Johar et al., 2009	<ul style="list-style-type: none"> <li>The only well documented registry in Saudi Arabia</li> <li>Data collected from patients seen in KFSH which is the major cleft center in Saudi Arabia</li> <li>Large total sample size (<math>n = 1317</math>)</li> </ul>	<ul style="list-style-type: none"> <li>Hospital based</li> <li>Prevalence was estimated and not mentioned in the article</li> <li>It is not the only referral center in Riyadh that provides OFC treatment nor is it a maternity hospital</li> <li>Heterogeneous sample from different regions. But, were mainly Saudis</li> <li>Prevalence was in living births only</li> </ul>
Borkar 1993	<ul style="list-style-type: none"> <li>Data collected from patients seen in the only tertiary Ministry Of Health hospital in the area</li> <li>It is mainly a prospective study</li> <li>The sample and population are homogeneous representing the region</li> </ul>	<ul style="list-style-type: none"> <li>Hospital based</li> <li>Not representative of the Saudi population as only set in one region of the country</li> <li>The age of patients included in the study was not mentioned</li> <li>The prevalence includes both syndromic and non-syndromic OFC</li> <li>Submucosal cleft was excluded.</li> <li>The prevalence was in living births only</li> </ul>
Al-Talabani et al., 1998	<ul style="list-style-type: none"> <li>The only maternity hospital in the region where 98% of the births take place</li> <li>It is a Prospective study</li> <li>Includes all births (live and stillbirths)</li> </ul>	<ul style="list-style-type: none"> <li>Heterogeneous sample, with different nationalities and culture</li> <li>Research objective involved assessing all major congenital malformation in UAE and did not focus on CLP</li> </ul>
Rajab and Thomas 2001	<ul style="list-style-type: none"> <li>Population based study</li> <li>Homogeneous sample with only Omanis</li> <li>Two sources of data collections were used to confirm the number of cases</li> <li>Large sample size (563)</li> </ul>	<ul style="list-style-type: none"> <li>Retrospective study</li> <li>The presence of home delivery in probably 20% of the cases</li> <li>It included both syndromic and non-syndromic OFC</li> </ul>
Patel 2007	<ul style="list-style-type: none"> <li>Population based study</li> <li>Included all health centers in the region</li> <li>Sample consists mainly of Omanis (96%) homogeneous</li> <li>Included all births (live and stillbirths)</li> </ul>	<ul style="list-style-type: none"> <li>The prevalence was in living births only</li> <li>Retrospective study</li> <li>The research objective involved assessing all major congenital malformation in Oman. Therefore, did not focus on CLP</li> </ul>
Al-Omari and Al-Omari 2004	<ul style="list-style-type: none"> <li>Data from the only two cleft centers in the country</li> <li>Representative of the population</li> <li>Large sample size (<math>n = 2146</math>)</li> </ul>	<ul style="list-style-type: none"> <li>Retrospective study</li> <li>It included both syndromic and non-syndromic OFC</li> <li>Prevalence was in living births only</li> <li>Submucosal clefts were excluded</li> </ul>
Aqrabawi 2008	<ul style="list-style-type: none"> <li>Prospective study Data collected from patients seen in the only maternity hospital in the city Amman</li> <li>The diagnosis of associated anomalies was standardized to all patients</li> </ul>	<ul style="list-style-type: none"> <li>Hospital based</li> <li>Can not be generalized to the population</li> <li>small sample size</li> <li>It includes both syndromic and non-syndromic OFC</li> <li>Family history was not clear</li> <li>The prevalence was in living births</li> </ul>

be less subject to error in these studies although it should be noted that their base population did not include stillbirths, which does cause some bias when comparing the prevalence reported in these studies to the WHO prevalence. However, this bias would probably be insignificant, since stillbirths account

for 1% of births in Oman (Patel, 2007) and 1.8% of births in Saudi Arabia (Ministry of Health, 2008). In addition, Rajab and Thomas (2001) reported that, according to the Ministry of Health of Oman, 20% of cases were home delivery. If these home births were not included in the denominator, then this

would lead to an overestimation of the OFC prevalence. Nevertheless, their result is supported by a similar regional study, which reported the same prevalence (Patel, 2007).

In Abu Dhabi, UAE, Al-Talabani et al. (1998) conducted a study in the only maternity hospital in the region. OFCs were recorded for both live and stillbirths. However, only 70% of this population was Arabs (Al-Talabani et al., 1998).

In Jordan, one study (Aqrabawi, 2008) reported a higher incidence of OFC (2.4/1000 live births) than the other (1.39/1000 live births) (Al-Omari and Al-Omari, 2004). The studies were carried out at the same hospital (KHMC), but the latter study had a larger sample size. Another peculiarity of the Aqrabawi study was that all CP cases were males, although all other studies report that CP occurs at a higher rate in females, in line with findings from other countries. Moreover, this study reported the highest prevalence of consanguinity (83%) in cases, but with no family history of CLP, which again contradicts the finding of other studies.

In the future, it might be possible to conduct a meta-analysis and explore factors associated with the variation of prevalence rates between studies. At present, the articles we found have insufficient numerical data for such an exercise to be meaningful.

#### 4.2. Prevalence of syndromes and associated anomalies

The reported prevalence of syndromes and associated abnormalities in these studies was unclear and lacked standardization. This lack of standardization could result from limited access to clinical geneticists immediately after the children were born, making it difficult to consistently record the exact diagnoses. Moreover, more minor anomalies could have been overlooked. Aqrabawi (2008) attempted to overcome this limitation by standardizing the methodology for diagnosis of all OFC cases born in the hospital by performing physical examination, 2D echocardiography, and renal ultrasound. While these data were classified according to whether they were associated with anomalies, the researcher did not record which results were syndromic.

Four of the studies reported associated anomalies in less than 30% of their OFC patients (Al-Johar et al., 2008; Al-Omari and Al-Omari, 2004; Borkar, 1993; Kumar et al., 1991). The Oman study reported the highest prevalence of associated anomalies, in 58.4% of all patients with OFC (Rajab and Thomas, 2001). This prevalence of associated anomalies is consistent with other investigations in different regions of the world that ranged from 21% to 63.4%, depending on the expertise, ascertainment, and definition of the associated anomalies (Mossey and Castilla, 2001; Shprintzen et al., 1985).

The most common associated anomaly reported was CHD. This association is supported by another study on congenital associated malformations in a sample of Jordanian patients with cleft lip and palate, which reported that 45.5% of the associated anomalies in OFC patients were CHDs (Rawashdeh and Abu-Hawas, 2008). This result is also consistent with other studies carried out in different parts of the world and the WHO global registry report (Milerad et al., 1997; Mossey and Castilla, 2001). On the other hand, the other Jordanian study (Aqrabawi, 2008) reported a higher rate of CHD among all cleft patients, which could be the result of using 2D-echocardiogram as a diagnostic aid for all newborn cleft patients.

Cleft palate was reported to have a higher prevalence of associated anomalies than other types of clefts. The prevalence rates of associated anomalies were 21.1% (Borkar, 1993) and 29.4% (Al-Bustan et al., 2002) in cleft palate cases, which is consistent with other reports in different parts of the world (Mossey et al., 2009; Rajab and Thomas, 2001; Rawashdeh and Abu-Hawas, 2008; Narchi and Kulaylat, 1997; Stoll et al., 2000).

#### 4.3. Consanguinity in families with cleft lip and palate

The types of consanguinity in Islamic and Arabian countries are first cousins, second cousins, double first cousins, or second cousins once removed. The prevalence of consanguineous marriages has been reported to be high (56%) in Saudi Arabia with approximately 60% of consanguineous marriages between first-degree cousins (El Mouzan et al., 2008; El-Hazmi et al., 1995). Some researchers have proposed consanguinity as a predisposing factor for OFC (Leitte and Koifman, 2009; Etahi et al., 2009). Therefore, it might be expected that there would be a high prevalence of OFC in the Saudi population. However, studies have reported the prevalence of OFC in children from consanguineous marriages to be lower than the general population (Al-Sahafi, 2010; Borkar, 1993; Kumar et al., 1991), which might indicate that consanguineous marriage alone is not a contributory factor to the prevalence of cleft lip and palate. Nevertheless, two of the studies reported a higher prevalence of first cousin consanguineous marriages within families with a history of cleft lip and palate than those reported in the general population. In these same studies, the presence of positive family history was reported in almost a quarter of the cleft patients (Al-Johar et al., 2008; Rajab and Thomas, 2001). This result is supported by Al-Bustan et al. (2002) who reported an insignificant, higher prevalence of positive family history in consanguineous marriages than in the general population (Al-Bustan et al., 2002). This report supports the claim that consanguineous parents with a family history of OFC have a higher chance of having children with OFC. Further investigations are needed to confirm these results.

## 5. Conclusions and recommendations

From this systematic review of the literature, we were unable to determine the prevalence of OFC in Saudi Arabia with certainty. Three studies of OFC prevalence in Saudi Arabia were identified that had a high degree of variation. KFSHRC showed the best registration of OFC in the country. It is important to continue this work (Al-Johar et al., 2009) by establishing a national registry of anomalies in Saudi Arabia. In the future, the reasons for the variation in OFC prevalence must be elucidated through more rigorous investigations of the epidemiology, geographic distribution, and etiology of cleft lip and palate in the country as a whole and in every region of the country using a standardized and collaborative research strategy.

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