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Biomedical Journal

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Original Article

Time trend of incidence rates of cleft lip/palate in Taiwan from 1994 to 2013

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ARTICLE INFO

Article history:
Received 20 August 2015
Accepted 5 October 2015
Available online 1 June 2016

Keywords:
Cleft lip
Cleft palate
Incidence
Prenatal diagnosis

ABSTRACT

Background: This study was to estimate the incidence rate of cleft lip and/or cleft palate (CL/P) in Taiwan from 1994 to 2013, and to assess the time trend over these years.
Methods: Retrospective data analysis was performed on records of all newborns with CL/P treated at Chang Gung Craniofacial Center, the only treatment center for CL/P in Taiwan, from 1994 to 2013. Three-year moving average rates were computed and linear regression was used to explore the annual average percentage change.
Results: From 1994 to 2013, 7282 newborns with CL/P were identified, corresponding to an annual rate of 1.48‰ (95% confidence interval (CI) = 1.45‰–1.52‰). There was a significant decline of rate of cleft lip with or without cleft palate (CL ± P) (−2.9% ± 0.2%, $p < 0.0001$) but slightly increase of rate of cleft palate (CP) only (+0.2% ± 0.07%, $p = 0.004$).
Conclusion: From 1994 to 2013, the annual rate of incidence of CL/P was 1.48‰ in Taiwan. The 2.9% annual decline of the rate was mainly from the CL ± P group, not the CP group.

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Peer review under responsibility of Chang Gung University.
http://dx.doi.org/10.1016/j.bj.2015.10.003

At a glance commentary

Scientific background on the subject

The incidence of orofacial clefts varied mainly from genetic and environmental reasons. Due to improving sonographic technology, prenatal diagnosis of cleft lip and/or cleft palate becomes accurate, and hence, the incidence is subject to alter.

What this study adds to the field

A decline in the incidence of orofacial clefts was observed from the group of cleft lip with or without cleft palate rather than the group of cleft palate in Taiwan between 1994 and 2013.

Cleft lip and/or cleft palate (CL/P) are the most common congenital craniofacial anomalies with an incidence of 1:700–1:1000. Multiple factors contribute to the development of cleft defect, including genetics, environments, and socioeconomics [1,2]. The prevalence is higher among Asians and people of native North American descent, followed by Caucasians, and least among Africans. The reported rate was 1.33/1000 live births for Asians, 1.30 for Chinese, 1.34 for Japanese, and 1.47 for other Asians [3]. In Taiwan, the annual incidence was reportedly 1.29/1000 in 1972 [4] and 1.12/1000 from 1980 to 1992 [5]. During 2002 and 2009, the overall annual prevalence of cleft deformities among 1,705,192 births was 1‰ for cleft lip with or without cleft palate (CL ± P) and 0.4‰ for cleft palate (CP) [6]. Environmental factors such as radiation, smoking, anticonvulsants, and alcohol consumption during pregnancy had been proposed as contributing factors to cleft development while folic acid was reported as a protective factor [7–9]. Low socioeconomic status, on the other hand, was found to be an indirect factor contributing to birth defect [10].

Prenatal diagnosis by sonography has become increasingly prevalent with improved accuracy [11]. The first sonography detection of cleft lip was reported in 1981 [12]. Major craniofacial anomalies can be identified by sonogram as early as 12 weeks of gestation [13]. The accuracy of transabdominal two-dimensional sonographic screening for orofacial clefts in a low-risk population ranged from 9% to 50% [14]. Three-dimensional sonography demonstrated enhanced accuracy of 100% for all clefts involving the primary palate and 86% of clefts involving secondary palate [14,15]. One study showed the rate of prenatal diagnosis of CL ± P increased from 11% to 50% from 1999 to 2008 [16]. While termination can only be performed for fetuses associated with severe anomalies, European studies reported that termination rate for solitary cleft lip and palate ranged from 3.3% to 9% [17].

In Taiwan, prenatal transabdominal ultrasound screening became readily accessible at a low cost since the establishment of National Health Insurance in 1995. Abortion is legally permitted before 24 weeks of pregnancy if the fetus has severe congenital anomalies or causes detrimental effect to the mother. Due to insufficient data on the birth prevalence and epidemiological characteristics of a facial cleft in Taiwan, we consider whether the prenatal diagnosis has an impact on the incidence of the facial cleft in Taiwan. In a culture where orofacial clefts are not well accepted and where there is not a stigma on abortion as compared to certain Catholic traditions, we suspect that the advent of orofacial cleft prenatal screening may influence the rate of abortion and the subsequent incidence of cleft deformity.

In this study, we investigated the change in the incidence of orofacial clefts in Taiwan from 1994 to 2013 during the enforcement of National Health Insurance since1995.

Methods

An institutional review board approval was obtained from the authors' institution. Data on all new births with cleft lip and palate in Taiwan from January 1994 to December 2013 were collected from two main centers in the hospitals: The Chang Gung Memorial Hospital (CGMH) in Linkou and the CGMH in Kaohsiung. The Plastic and Reconstructive Surgery Department in both hospitals held the only Craniofacial Center where all patients with cleft deformity were referred. Data on the prenatal diagnosis rate of cleft lip and palate were partially available from chart review of all patients undergoing treatment at CGMH in Linkou under one physician between January 2009 and December 2012. As CP was rarely diagnosed prenatally, data on the frequency of prenatal diagnoses were concentrated on CL ± P.

Live birth data was obtained from the Department of Statistics at the Ministry of Interior in Taiwan. The incidence of oral clefts in the present study was based on live births. Rates were calculated as the numbers of event in 1994–2013 dividing by the live births of the same year. To minimize fluctuation of the annual rates, 3-year moving average rates were computed [18]. The 95% CIs of the rate were calculated assuming a Poisson distribution. Linear regression of the rate has been used to look at annual average percentage change [19]. Interaction between time and group (CL ± P vs. CP) in the linear regression was added to examine whether there is difference in the slopes between two groups. $P < 0.05$ was taken to be statistically significant.

Results

A total of 7282 new patients with CL/P from 1994 to 2013 were identified. The estimate of the birth incidence was based on 4,912,739 total live births, according to the Department of Statistics at the Ministry of Interior in Taiwan. The annual incidence for cleft births over the 20-year period was 1.48/1000 (95% CI = 1.45–1.52), or 1/675 live births [Table 1]. Linear regression revealed on the 3-year moving average rates that there was a significant decline of CL ± P rate (−2.9% ± 0.2%, $p < 0.0001$) but slightly increase of CP rate (+0.2% ± 0.07%, $p = 0.0040$) [Fig. 1].

The prenatal diagnostic rate of CL ± P from January 2009 to December 2012 was found to be 73% among all children with varied severity of cleft deformity undergoing treatment by the senior author. A total of 148 children were seen, among which 108 confirmed the diagnosis from the prenatal ultrasound.