Hospitalisations up to adulthood for children born with orofacial clefts

ABSTRACT

Aim: To compare hospital admissions from infancy to adulthood, between children born with orofacial clefts (OFC) and those without OFC.

Methods: Cohort study using record-linked administrative datasets. Participants included all children liveborn in Western Australia (WA) 1980-2010 and diagnosed with OFC, frequency matched by year of birth to randomly selected liveborn children without OFC. We calculated rate ratios (RR) of hospital admission, number and reason of admissions, cumulative length of stay, for each cleft type (cleft lip only [CLO], cleft lip and palate [CLAP], cleft palate only [CPO], no OFC) and by age period (infancy, pre-school, primary and high school ages and early adulthood)

Results: Overall, 1396 children were diagnosed with an OFC and compared with 6566 children without OFC. Individuals born with OFC were up to three times more likely to be admitted to hospital, had more admissions and longer cumulative length of stay in all age periods. Children with OFC were also more likely to be admitted for ear and digestive system conditions (RR up to 30 and 6 times higher respectively). Children with CLAP and CPO were more likely to be admitted for respiratory conditions (RR 1.3 to 2.0) and children with CPO were six times more likely to be admitted for care for other congenital anomalies.

Conclusions: Throughout childhood, individuals born with OFC were more likely to be admitted, and had more hospitalisations than those without OFC. Children born with CLAP or CPO had a higher hospitalisation burden than children born with CLO.

Key words: cleft lip, cleft palate, hospitalisations, medical record linkage, Western Australia

WHAT IS ALREADY KNOWN:

- Burden of all-cause hospital admissions for children with clefts is greater than for children without clefts at younger ages
- Hospitalisation is greater for children with cleft lip and palate or cleft palate only than for children with cleft lip only

WHAT THIS STUDY ADDS:

- Difference in admissions between those with and without clefts is greatest in infancy and younger ages, and although continues to adulthood, narrows as age increases
- Burden of hospitalisations is related to cleft management and admissions associated with respiratory, middle ear and dental conditions, and other congenital anomalies
- Number of admissions has remained constant, but length of stay in hospital has declined in more recent years

INTRODUCTION

Orofacial clefts (OFC) occur in around 1 in 700 births.[1-3] Children born with OFC need multidisciplinary care from birth to adulthood, including surgery, speech therapy, general dentistry and orthodontics.[4]

Few population-based studies have investigated the use of hospital services for children with OFC. Increased admission rates and duration of time in hospital, accompanied by substantial higher hospitalisation costs have been described for children with OFC compared to children without OFC,[5-7] but these studies have focused on all admissions, particularly those in the first one or two years of life. More recently, Fitzsimons et al[8] described admissions from all causes as well as those directly related to OFC in the first two years of life. Only one study has investigated admissions beyond childhood by modelling the probability of admission and length of stay over the life-span for those with and without OFC.[9] No study has described in detail, all admissions until adulthood.

Using linked population-based data from Western Australia (WA), we describe the hospitalisation experience of individuals born with OFC by comparing their likelihood and number of admissions, time spent in hospital and reasons for admission, with individuals not born with OFC, from infancy to adulthood. This information is important for families with children born with OFC, as well as for health professionals, planners and policy makers.

METHODS

Data sources

We used record-linked data from five data collections in WA. The WA Register of Developmental Anomalies (WARDA), is a population-based statutory system of congenital anomalies, with multiple sources of ascertainment of structural and functional anomalies diagnosed up to six years of age. The Midwives Notification System (MNS) is a legislated surveillance system covering all births in WA of >20 weeks gestation or 400g birthweight. The Birth and Death Registries collect data on all births and deaths registered in WA. The Hospital Morbidity Data System is a census of all public and private inpatient admissions in WA. Individual's records from these sources were linked by the WA Data Linkage System using probabilistic matching.

Study population

All infants liveborn in WA between 1980 and 2010, with OFC were identified from the WARDA using the BPA-ICD9 codes for cleft palate only (CPO) (74900-74909), cleft lip only (CLO) (74910-74919) and for cleft lip and palate (CLAP) (74920-74927, 74929). If other congenital anomalies, were recorded as well as OFC, these infants were defined as having an additional anomaly. Cases of OFC without other anomalies were defined as isolated. A comparison cohort, frequency matched on year of birth and not diagnosed with OFC, was randomly selected from liveborn infants recorded in the MNS.

Hospital admissions

We received all hospital admission records from 1 January 1980 to 31 December 2012 for study participants. Hospital birth records were provided only for infants requiring admission; hospital birth records for healthy infants were not provided. Where consecutive admission records indicated hospital transfers or a change in the type of admission, these records were merged into one admission. Length of hospital stay was calculated as the time between admission and discharge. Where admission and discharge occurred on the same day, the duration of hospital stay was considered to be 0.5 day, to reflect some period of hospitalisation.[10]

From 1980 to 1987, diagnoses were coded according to ICD-9 and procedures using the International Classification of Procedures in Medicine (1978). From January 1988 to June 1999, diagnoses and procedures were coded according to ICD-9 CM, and since July 1999, ICD-10-AM and the Australian Classification of Health Interventions have been used to code diagnoses and procedures respectively. To allow comparisons across the study period, all diagnoses and procedures were mapped to ICD-9-CM codes.

Admissions were defined as cleft-related or not (Table 1), after discussion with cleft treatment experts (DG, WM) and included both surgical and non-surgical admissions. We defined reasons for non-cleft-related admissions using the principal diagnosis categorised according to ICD-9 chapter headings. As admissions for middle ear related conditions are common among infants with OFC,[11] we separated admissions for diseases of the ear from diseases of the nervous system. Readmissions within 28 days were excluded from the analyses describing reasons for non-cleft-related admissions.

Analyses

We conducted analyses by cleft type (CLO, CLAP, CPO, no OFC) in five age periods: infancy (up to 1 year), pre-school (1-<5 years), primary school (5-<12 years), high school (12-<18 years), and early adulthood (18-<25 years). As admissions may not occur randomly across an age period (for example, cleft repair and revision follows a protocol, with treatment planned at specified ages), children without complete follow up over a whole period were excluded from analyses for those age periods. Complete years of follow up were not available for all children in all age periods if their birth was too recent for complete follow up, or if they died during the age period.

For each child in each age period, we compared the number of admissions and cumulative length of stay (cLOS) between children with and without OFC, and between those with and without additional anomalies. Admission rates (number of children admitted per number children who could potentially be admitted) for each period, and rate ratios were compared between children with and without OFC. For children with follow-up to 18 years, we examined the number of cleft-related surgical admissions after the primary repair. To examine possible changes in admission practices over time, we compared the number of admissions and cLOS between individuals with admissions in the 2000s to those with admissions in 1980-1999.

We used the Wilcoxon Sum Rank test to compare differences in numbers of admissions and cLOS, and calculated 95% confidence intervals around relative rates of admission. To avoid the potential for identifying individuals, we suppressed results with ≤ 10 people. Statistical analyses were conducted using SAS 9.3[12] and Epi Info 7.[13]

Members of CleftPALS WA, the support group for families with members affected by OFC, provided advice for this study. The study protocol was approved by the WA Department of Health Human Research Ethics Committee and the WA Aboriginal Health Ethics Committee. These approvals were recognised by the University of Sydney's Human Research Ethics Committee.

RESULTS

Of the 1396 children born with OFC between 1980 and 2010 and alive at one year, 345 were born with CLO, 412 with CLAP, and 639 with CPO (Table 2). The comparison group included 6566 infants without a cleft. The majority of clefts involving the lip were unilateral

(77.4% of those with CLO, 68.0% of children with CLAP; 6.1% and 25.0% were bilateral respectively). Most children with CPO (77.8%) had clefts involving the hard or soft palate or both. Nearly all (98.8%) children with OFC and 69.8% of the comparison group were hospitalised at least once before their 25th birthday. Children who died during an age period were not included in analyses for that period (132 children with OFC and 63 without OFC; 186 and 127 admissions respectively).

The burden of hospitalisation, including the proportion of children admitted, number of admissions, and cLOS was considerably greater for children with OFC than for children without OFC, was highest in infancy and between ages 5-<12 years, and this disparity continued up until adulthood (Table 3). Generally, children with CLAP or CPO had more admissions and longer cLOS than children with CLO, and children with CLAP had the highest rates of admission in every age period.

In their first year, nearly all children diagnosed with OFC were admitted to hospital, compared with 36% of children without OFC. Primary cleft repair (Table 4) usually occurred in the first year and was a major contributor to admissions in this period. However, the age at repair for children with a submucous cleft palate was significantly older (approximately 3 years 9 months) ($p< 0.0001$). Admissions for non-surgical cleft-related care (such as nutritional needs, social work, breathing difficulties) were also common in the first year, accounting for 35.1% of cleft-related admissions.

In each age period up to 12 years, children with and without OFC and with an additional anomaly had more admissions and longer cLOS in hospital compared to children with isolated OFC, or with no OFC respectively. In adulthood (18-<25 years), around 50% of adults with or without OFC were admitted to hospital with the median number of admissions and cLOS in hospital similar for all groups. Those with CLAP however, were more likely to be admitted (Table 3).

For children with complete follow-up until 18 years, most with CLO or CPO (53.0% and 66.0% respectively) had no further cleft-related surgery after primary repair. Another 31.3% and 23.0% respectively had one additional surgical admission, the remainder having more. Among children with CLAP, 9.5% had no further surgical admissions, 14.5% had one, and the majority (60.9%) had 2-4 additional surgical admissions.

Children admitted in 2000-2012, with or without OFC, generally spent less time in hospital, despite having similar numbers of admissions, compared to admissions during 1980-1999 (Table 5).

After excluding cleft-related admissions, there were marked differences in the rates and reasons for admission for those with OFC compared to those without (Table 6). Among children with OFC, the pattern and relative rate of admission was similar between those with CLAP and CPO, with their relative rate of admission being higher than for those with CLO. For all children with OFC, particularly those with CLAP and CPO, admission rates for ear problems (92.6% were middle ear conditions) were 10-40 times higher, and digestive system problems (70.6% admissions after one year of age were dental diagnoses) were 4-6 times higher up to the age of 12 years and twice as high in the 12-<18 year age-period. Admissions for respiratory conditions were 1.3-2.0 times more likely for children with CLAP or CPO (but not CLO) in infancy and the pre-school period. In the same age periods, children born with CPO were around six times more likely to be admitted for treatment of another congenital anomaly compared to admissions for congenital anomalies in the comparison group. In the oldest age period, admission rates for the most common diagnostic categories amongst those with OFC were no different to those for adults without OFC.

DISCUSSION

This is the first study to describe in detail, all hospital admissions up to adulthood, for children born with OFC. The increased likelihood and number of admissions, and longer cLOS for individuals with OFC highlight the disparity in burden of hospitalisation for these children. The use of hospital services was highest in the infancy and primary school aged years but continued up until adulthood. Children with OFC and an additional anomaly had more admissions than children with isolated OFC up until age 12 years. cLOS was also longer for children with an additional anomaly during infancy (all cleft types), pre-school (children with CLAP and CPO), and in the primary school period for children with CPO.

Admissions in the early years for children with OFC were similar to those found in other population-based studies,[5, 8] although differences in age groups hinder direct comparisons. Only one other population-based study has investigated differences in hospitalisations into adulthood and reported that individuals with OFC had greater use of hospital services than

individuals without OFC in all age groups up to 60 years old, and that differences decreased with age.[9]

Most other population-based studies of hospital admissions for children with OFC have investigated all-cause admissions.[5-7, 9] Only one other study has distinguished admissions directly related to cleft management from other admissions.[8] In that study, surgical admissions related to the face, mouth, palate, pharynx nose and middle ear were designated cleft admissions; non-surgical cleft-related admissions (for interventions such as nutrition and counseling) were excluded from this category, thereby under-estimating cleft-related hospitalisations.[8]

Reasons for admissions for children without OFC were similar to those reported in a large population-based study, also from WA, comparing admissions between children with and without congenital anomalies.[10] In our study, children with OFC consistently had higher admission rates for respiratory, ear, and digestive system conditions. The higher rates for respiratory admissions in infancy and in the pre-school period reflect findings from another population-based study of admissions for acute lower respiratory infections (ALRI), where children born with CLAP or CPO had over twice the risk of admission for an ALRI during the first two years of life compared with children without congenital anomalies.[14] High prevalence of middle ear conditions are well documented,[11, 15] but no population-based comparisons for middle ear and dental conditions have been reported. Even though our admission rates were high, they may be under-estimated if procedures for other conditions (such as procedures for middle ear conditions) were also performed during admissions for cleft management. Higher relative rates of admission for children with OFC are also possible if they are more likely to be admitted for these other conditions (and less likely to be treated out of hospital) than children without OFC.

The distinction between cleft-related admissions and other admissions may however, be artificial as anatomical differences in children with OFC may increase vulnerability to these conditions. Children with cleft palate have impaired Eustachian tube function increasing susceptibility to otitis media with effusion.[11, 16] Increased need for dental services may be explained by disruption at the cleft site affecting tooth development,[17] making good oral hygiene difficult and increasing susceptibility to dental caries,[18] or requiring extraction as part of the treatment. Similarly the increased risk of respiratory infections for children with

CLAP or CPO may be due to anatomical defects that increase the risk of pathogens entering the upper respiratory tract and lung, and surgery under anaesthetic (for which children with OFC have at least one admission during their first year) may also contribute.[14] While the distinction between cleft and non-cleft-related admissions may be debatable, it highlights the additional hospital use for children with OFC, above that directly related to cleft management.

Our study covering 25 years, using linked data from 1980, and with OFC status defined using a population-based congenital anomaly register with active surveillance, enabled comparisons in all admissions between those born with OFC, and those without. Completeness of registrations of individuals with OFC is estimated to be 100%[19] and with diagnoses up to six years of age, children with all cleft types were included. The quality of our data is high with linkage proportions >99%.[20]

Diagnosis and procedure coding changed twice during the study period, potentially affecting accuracy.[21] Although coding became more detailed in later versions, we were limited to the broader categories defined by the earlier coding systems. There are no validation studies evaluating the accuracy of diagnoses and procedures for admissions related to cleft management, but in the WA hospital morbidity data, procedures are more accurately recorded than diagnoses, and major procedures and diagnoses are likely to be identified.[22] Although we potentially misclassified some cleft and non-cleft admissions, all-cause admission analyses would not have altered.

Excluding infants who died after the beginning of an age period may have under-estimated the use of hospital services in both those with and without OFC, if they were relatively higher users of hospital services. However, the small number of children who died seems unlikely to have affected our results. We consider that children who were born too recently to contribute data over a complete age period were likely to have similar hospital use to those included in those analyses, and their exclusion was unlikely to bias results. We could not determine how many participants moved out of state during the study period, but migration out of WA was low (around 2.8% in 2003 and 2004).[23]

Our study provides a longitudinal, population-based description of all hospital admissions for individuals with all cleft types, and highlights their disproportionate use of hospital services

compared to individuals without OFC. The extent of their hospital use may have implications for children's development as well as a large impact on their families. This information provides a foundation for counseling families about expectations for admissions, and is also useful for health care providers and service planners.

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Table 1. Cleft related admissions

Cleft related admissions included all admissions where the main diagnosis was attributed to an orofacial cleft (OFC) or where specific procedure codes indicated cleft repair (cleft lip or cleft palate repair, cleft palate revision), regardless of the main diagnosis code. In addition, other admissions were also defined as cleft related where plastic procedures to the mouth, nose, palate or pharynx, or orthognathic surgery were conducted (similar to definitions used by Fitzsimons et al[8]). Other admissions where non-plastic procedures to the nose or mouth and pharynx were recorded were also regarded as cleft admissions, but only if they occurred in admissions for children with diagnostic codes indicating scars, nose anomalies, lip conditions, or dentofacial anomalies. Unlike Fitzsimons et al, admissions where procedures to the middle ear were performed, or any procedures to nose or mouth region, were not classified as cleft related admissions unless they were performed in an admission meeting the criteria above. Any admissions where the principal diagnosis indicated injury (unrelated to complications of surgical or medical care) were not regarded as cleft related admissions. More detail is provided in the table below.

Criteria defining cleft related admissions[†]

Principal diagnosis code	Procedures
OFC	Procedures to mouth face, or for grafts, scar or flap
OFC	No procedures related to cleft surgery
Any	Cleft lip or palate repair or revision, orthognathic surgery, procedure to the uvula, bone graft, other plastic procedures to palate, plastic procedures to the pharynx, nose, or mouth
Deviated septum, scars, anomalies of nose, lip diagnoses	Non-plastic procedures to the nose
Diagnoses of scars, anomalies of nose, lip diagnoses, dentofacial anomalies	Non-plastic procedures to the mouth

[†] Admissions where main diagnosis was coded to injury (other than a medical complication) were not regarded as cleft admissions.

Admission for primary repair of the cleft

The admission for primary repair of the cleft was defined as the first admission with a procedure code for cleft repair. Additional admissions for primary repair of the palate were identified where, up to one year old, a procedure code indicating a revision of the cleft was recorded but no primary repair code.

Table 2. Description of children included in cohort,[†] by type of cleft

	Cleft type							
	CLO		CLAP		CPO		No cleft	
	n	(%)	n	(%)	n	(%)	n	(%)
Number of children	345		412		639		6566	
Sex - male	218	(63.2)	267	(64.8)	278	(43.5)	3402	(51.8)
Diagnosis of another anomaly[‡]	46	(13.3)	91	(22.1)	335[§]	(52.4)	330	(5.0)
Decade of birth								
1980s	108	(31.3)	148	(35.9)	155	(24.3)	2064	(31.4)
1990s	98	(28.4)	119	(28.9)	212	(33.2)	2089	(31.8)
2000s	139	(40.3)	145	(35.2)	272	(42.6)	2413	(36.8)
N with a hospital admission record[¶]	338	(98.0)	411	(99.8)	630	(98.6)	4586	(69.8)

[†] Born alive in WA 1980-2010, alive at 1 year of age. [‡] Another congenital anomaly, including a diagnosis of chromosomal anomalies and syndromes. [§] Includes 141 children diagnosed with Robin Sequence. [¶] For admissions between 1980-2012 and up to 25th birthday; admissions at birth for normal healthy babies were not provided by data custodians and therefore are not included.

Table 3. Hospital admissions for children born in WA 1980-2010, with admissions 1980-2012, by cleft type and age period

	CLO		CLAP		CPO		No OFC	
Birth up to 1 year								
N infants[†]	345		412		639		6566	
N admitted (% of infants)	319	(92.5)	409	(99.3)	559	(87.5)	1969	(30.0)
RR admission (95% CI)	3.08	(2.94, 3.23)	3.31	(3.19, 3.44)	2.92	(2.78, 3.06)	Ref	
N with cleft admission (% of infants)	299	(86.7)	406	(98.5)	453	(70.9)	-	
N with non-cleft admission (% of children)	114	(33.0)	176	(42.7)	333	(52.1)	1969	
N with admission in first week (% of children)	131	(38.0)	382	(92.7)	414	(64.8)	955	(14.5)
N admissions	599		1462		1479		2848	
N cleft-related admissions (% of admissions)	401	(66.9)	1137	(77.8)	732	(49.5)		
Median N admissions[‡] (P10, P90)	2[*]	(1, 3)	3[*]	(2,5)	2[*]	(1, 4)	1	(1,2)
No additional congenital anomaly	1	(1, 3)	3	(2, 5)	2	(1, 3)	1	(1, 2)
Additional congenital anomaly	2[**]	(1, 4)	4[**]	(2, 6)	2[**]	(1, 5)	2[**]	(1, 4)
Median cumulative LOS[‡] (P10, P90)	7[*]	(2, 21)	19.5[*]	(9.5, 49)	13[*]	(3.5, 54)	4	(0.5, 17)
No additional congenital anomaly	7	(2, 16)	19	(9.5, 40)	8	(3, 18.5)	4	(0.5, 16)
Additional congenital anomaly	10.25[**]	(3, 43)	25[**]	(10, 82)	20[**]	(6, 79)	6[**]	(0.5, 41)
1 up to 5 years								
N children[†]	309		367		549		5878	
N admitted (% of children)	167	(54.0)	315	(85.8)	453	(82.5)	2013	(34.2)
RR admission (95% CI)	1.58	(1.42, 1.76)	2.51	(2.37, 2.65)	2.41	(2.29, 2.54)	Ref	
N with cleft admission (% of children)	33	(10.7)	128	(34.9)	126	(23.0)		
N with non-cleft admission (% of children)	156	(50.5)	297	(80.9)	434	(79.1)	2013	
N admissions	458		1053		1581		3590	
N cleft-related admissions (% of admissions)	39	(8.5)	164	(15.6)	155	(9.8)		
Median N admissions[‡] (P10, P90)	2[*]	(1, 5)	3[*]	(1, 6)	2[*]	(1, 7)	1	(1, 3)
No additional congenital anomaly	1	(1, 5)	2	(1, 5)	2	(1, 4)	1	(1, 3)
Additional congenital anomaly	2.5[**]	(1, 9)	4[**]	(2, 9)	3[**]	(1, 9)	2[**]	(1, 5)
Median cumulative LOS[‡] (P10, P90)	2[*]	(0.5, 11.5)	3[*]	(0.5, 23)	3[*]	(0.5, 21)	1.5	(0.5, 8)
No additional congenital anomaly	1.5	(0.5, 9)	2.5	(0.5, 18)	1.5	(0.5, 14)	1	(0.5, 7)
Additional congenital anomaly	2.5	(0.5, 25.5)	4[**]	(1, 33.5)	4.5[**]	(0.5, 29)	2[**]	(0.5, 18)

5 up to 12 years								
N children[†]	220		276		382		4367	
N admitted (% of children)	136	(61.8)	246	(89.1)	273	(71.5)	1426	(32.7)
RR admission (95% CI)	1.89	(1.69, 2.12)	2.73	(2.57, 2.90)	2.19	(2.03, 2.36)	Ref	
N with cleft admission (% of children)	53	(24.1)	213	(77.2)	48	(12.6)		
N with non-cleft admission (% of children)	120	(54.5)	199	(72.1)	259	(67.8)		
N admissions	292		882		841		2484	
N cleft-related admissions (% of admissions)	64	(21.9)	306	(34.7)	66	(7.9)		
Median N admissions[‡] (P10, P90)	2[*]	(1, 4)	3[*]	(1, 7)	2[*]	(1, 6)	1	(1, 3)
No additional congenital anomaly	2	(1, 4)	3	(1, 6)	2	(1, 5)	1	(1, 3)
Additional congenital anomaly	3[**]	(1, 8)	4[**]	(1, 8)	3[**]	(1, 6)	2[**]	(1, 6)
Median cumulative LOS[‡] (P10, P90)	2.5[*]	(0.5, 9)	6.5[*]	(2.5, 19)	2.5[*]	(0.5, 10.5)	1	(0.5, 6.5)
No additional congenital anomaly	2.25	(0.5, 8.5)	6.5	(2.5, 16)	2	(0.5, 8)	1	(0.5, 6)
Additional congenital anomaly	3.5	(1, 17.5)	6	(2.5, 29.5)	2.75[**]	(0.5, 17)	2[**]	(0.5, 21)
12 up to 18 years								
N children[†]	153		195		242		3030	
N admitted (% of children)	67	(43.8)	142	(72.8)	119	(49.2)	1020	(33.7)
RR admission (95% CI)	1.30	(1.08, 1.57)	2.16	(1.96, 2.39)	1.46	(1.27, 1.68)	Ref	
N with cleft admission (% of children)	19	(12.4)	70	(35.9)	18	(7.4)		
N with non-cleft admission (% of children)	58	(37.9)	119	(61.0)	116	(47.9)	1020	
N admissions	120		324		275		1771	
N cleft related admissions (% of admissions)	20	16.7	105	32.4	22	8.0		
Median N admissions[‡] (P10, P90)	1	(1, 4)	2[*]	(1, 4)	2[*]	(1, 5)	1	(1, 3)
No additional congenital anomaly	1	(1, 4)	2	(1, 4)	1.5	(1, 3)	1	(1, 3)
Additional congenital anomaly	[§]		2	(1, 6)	2	(1, 5)	1	(1, 3)
Median cumulative LOS[‡] (P10, P90)	1	(0.5, 9)	2[*]	(0.5, 8)	1.5[*]	(0.5, 10)	1	(0.5, 8)
No additional congenital anomaly	1	(0.5, 9)	2	(0.5, 7.5)	1.5	(0.5, 7)	1	(0.5, 8)
Additional congenital anomaly	[§]		1.5	(0.5, 13.5)	2	(0.5, 10)	1	(0.5, 15.5)
18 up to 25 years								

N adults[†]	74		116		121		1609	
N admitted (% of adults)	33	(44.6)	71	(61.2)	67	(55.4)	799	(49.7)
RR admission (95% CI)	0.90	(0.69, 1.16)	1.23	(1.06, 1.44)	1.12	(0.94, 1.32)	Ref	
N with cleft admission (% of children)	[§]		32	(27.6)	[§]			
N with non-cleft admission (% of children)	32	(43.2)	57	(49.1)	65	(53.7)	799	(49.7)
N admissions	87		181		156		1822	
N cleft related admissions (% of admissions)	[§]		51	(28.2)	[§]			
Median N admissions[‡] (P10, P90)	1	(1, 5)	2	(1, 5)	2	(1, 5)	2	(1, 4)
No additional congenital anomaly	1	(1, 5)	2	(1, 4)	1	(1, 4)	2	(1, 4)
Additional congenital anomaly	[e]		[§]		2	(1, 5)	2	(1, 4)
Median cumulative LOS[‡] (P10, P90)	1	(0.5, 11)	2	(0.5, 11)	3	(0.5, 14)	2	(0.5, 11)
No additional congenital anomaly	2	(0.5, 9)	2	(0.5, 10)	2	(0.5, 11)	2	(0.5, 11)
Additional congenital anomaly	[§]		[§]		4	(0.5, 15.5)	1.5	(0.5, 15)

[†] Denominator includes children with complete follow up over the whole period. [‡] For individuals admitted to hospital. [§] ≤ 10 children admitted, data suppressed. [*] $p<0.05$ compared to individuals without clefts. [**] $p<0.05$ for comparison between individuals with isolated clefts and additional anomalies; or in the comparison group, between those with no anomaly and those with a congenital anomaly. CI, confidence interval; LOS, length of hospital stay (days); P10, P90, 10th and 90th percentiles; Ref, reference group; RR, relative rate.

Table 4. Admission for primary repair of cleft

	Cleft type			
	CLO	CLAP		CPO[†]
N children[‡]	345	412		589
Type of cleft repair	lip	lip	palate	palate
N with primary repair identified	307	402	378	507
Median age (days)	102	92	279	286
cleft of hard and, or soft palate				282[*]
submucous CPO				1368[*]
Age by which 90% children had repair done (days)	202	128	385	525
cleft of hard and, or soft palate				383
submucous CPO				2372
Median LOS (days)	6	7	7	5[**]
LOS (days) for 80% of children[§]	2-8	2-11	2-13	2-12

[†] Excludes infants born with bifid uvula only (n=50). [‡] Alive at 1 year old. [§] LOS for 80% of children (10th and 90th percentiles). [*] difference in age $p < 0.0001$. [**] difference in LOS $p = 0.002$ between infants with isolated CPO (4 days) and those with CPO and additional anomaly (6 days). No significant difference in LOS between infants with isolated or additional anomalies for infants with CLO or CLAP. LOS, length of stay.

Table 5. Median number of admissions and cumulative length of stay for children born in WA 1980-2010, for admissions 1980-1999 and 2000-2012, by cleft type and age period[†]

	CLO				CLAP				CPO				No OFC			
	1980-99		2000-12		1980-99		2000-12		1980-99		2000-12		1980-99		2000-12	
Birth up to 1 year																
N admissions (P10, P90)	2	(1,3)	2	(1,3)	3	(2,5)	3	(3,5)	2	(1,4)	2	(1,4)	1	(1,2)	1	(1,2)
cLOS (P10, P90)	8	(5,20)	5***	(1.5,20)	25	(13,57)	14***	(8.5,27)	14	(5.5,55)	10***	(3,50)	5	(1,19)	3***	(0.5,17)
Primary repair admission																
LOS (P10, P90)	7	(5,9)	2***	(1,5)	lip: 8	(5,12)	3***	(2,5)	8	(5,13)	3***	(2,5)				
					palate: 9	(5,13)	3***	(2,5)								
1 up to 5 years																
N admissions (P10, P90)	2	(1,4)	2	(1,5)	3	(1,6)	2*	(1,5)	3	(1,7)	2	(1,6)	1	(1,3)	1	(1,3)
cLOS (P10, P90)	2.5	(0.5,10.5)	1*	(0.5,8.5)	6.5	(0.5,26.5)	1.5***	(0.5,4.5)	5	(0.5,26.5)	1.5***	(0.5,10)	1.5	(0.5,9)	1***	(0.5,5)
5 up to 12 years																
N admissions (P10, P90)	2	(1,4)	2	(1,4)	3	(1,6)	3	(1,6)	2	(1,6)	2	(1, 4)	1	(1,3)	1	(1,3)
cLOS (P10, P90)	3	(0.5,12)	1.5	(0.5, 5.5)	9	(3.5,22)	4.5***	(1,10)	2.5	(0.5,10)	2*	(0.5,5.5)	1.5	(0.5,7)	1***	(0.5,5)
12 up to 18 years																
N admissions (P10, P90)	2	(1,4)	1	(1,3)	2	(1,5)	1	(1,3)	1	(1,3)	2	(1,5)	1	(1,3)	1	(1,3)
cLOS (P10, P90)	1	(0.5,16.5)	1.5	(0.5,8)	2	(0.5,7)	1.5	(0.5,5)	1	(0.5,6.5)	1.5	(0.5,7.5)	1	(0.5,7)	1**	(0.5,7.5)

[†] 18-<25 years: too few (≤10) individuals with admissions in 1980-99 in each cleft group to enable comparison between admission periods. * $p < 0.05$, ** $p<0.01$, *** $p< 0.001$ for decline from 1980-1999 to 2000-2012, Wilcoxon Sum Rank test. cLOS, cumulative length of stay; P10 P90, 10th and 90th percentiles.

Table 6. Most common principal diagnoses[†] for admissions (not cleft-related and not readmissions) for children born in WA 1980-2010 with admissions 1980-2012, by cleft type and age period[‡]

	CLO			CLAP			CPO			No OFC		
Birth up to 1 year												
N infants	345			412			639			6566		
N admissions	138			202			506			2566		
N infants w admission	104			148			309			1969		
Principal diagnosis	*N*	*%*	*RR (95%CI)*		*N*	*%*	*RR (95%CI)*		*N*	*%*	*RR (95%CI)*		*N*	*%*
Perinatal	43	12.5	0.98 (0.74, 1.31)	Respiratory	36	8.7	1.54 (1.11, 2.13)	Other CA	84	13.1	6.50 (5.00, 8.42)	Perinatal	833	12.7
Respiratory	15	4.3	0.77 (0.46, 1.27)	Ear	32	7.8	18.89 (11.43, 31.22)	Perinatal	78	12.2	0.96 (0.77, 1.20)	Respiratory	373	5.7
Factors	13	3.8	1.05 (0.61, 1.81)	Symptoms	21	5.1	1.41 (0.91, 2.17)	Respiratory	73	11.4	2.01 (1.59, 2.55)	Symptoms	238	3.6
Other CA	§			Infection	18	4.4	1.35 (0.85, 2.17)	Symptoms	61	9.6	2.63 (2.01, 3.45)	Factors	236	3.6
Symptoms	§			Factors	18	4.4	1.22 (0.76, 1.94)	Ear	37	5.8	14.08 (8.63, 22.97)	Infection	212	3.2
Ear	§													
1 up to 5 years												
N children	309			367			549			5878		
N admissions	335			779			1239			3285		
N children w admission	152			296			433			2010		
Principal diagnosis	*N*	*%*	*RR (95%CI)*		*N*	*%*	*RR (95%CI)*		*N*	*%*	*RR (95%CI)*		*N*	*%*
Digestive	51	16.5	3.33 (2.53, 4.39)	Ear	225	61.3	12.09 (10.54, 13.87)	Ear	289	52.6	10.38 (9.06, 11.90)	Respiratory	680	11.6
Respiratory	43	13.9	1.20 (0.90, 1.60)	Digestive	112	30.5	6.16 (5.09, 7.46)	Digestive	119	21.7	4.38 (3.60, 5.32)	Injury	392	6.7
Ear	42	13.6	2.68 (1.98, 3.63)	Respiratory	57	15.5	1.34 (1.05, 1.72)	Respiratory	114	20.8	1.80 (1.50, 2.14)	Ear	298	5.1
Injury	23	7.4	1.12 (0.74, 1.67)	Injury	42	11.4	1.72 (1.27, 2.32)	Symptoms	69	12.6	3.04 (2.36, 3.91)	Digestive	291	5.0
Infection	21	6.8	1.54 (1.00, 2.37)	Symptoms	28	7.6	1.86 (1.27, 2.69)	Other CA	62	11.3	6.03 (4.48, 8.14)	Infection	259	4.4
5 up to 12 years														

	CLO			CLAP			CPO			No OFC		
N children		220			276			382			4367	
N admissions		206			545			706			2308	
N children w admission		119			196			259			1426	
Principal diagnosis	N	%	RR (95%CI)		N	%	RR (95%CI)		N	%	RR (95%CI)		N	%
Digestive	63	28.6	4.39 (3.46, 5.56)	Ear	119	43.1	11.14 (9.12, 13.62)	Ear	152	39.8	10.28 (8.48, 12.47)	Respiratory	416	9.5
Respiratory	27	12.3	1.29 (0.89, 1.86)	Digestive	114	41.3	6.33 (5.29, 7.58)	Digestive	100	26.2	4.01 (3.28, 4.91)	Injury	378	8.7
Injury	22	10.0	1.16 (0.77, 1.74)	Injury	34	12.3	1.42 (1.02, 1.98)	Respiratory	47	12.3	1.29 (0.97, 1.71)	Digestive	285	6.5
Ear	13	5.9	1.53 (0.88, 2.64)	Factors	26	9.4	5.96 (3.86, 9.20)	Injury	47	12.3	1.42 (1.07, 1.89)	Ear	169	3.8
Skin	§			Respiratory	24	8.7	0.91 (0.62, 1.35)	Symptoms	25	6.5	2.22 (1.46, 3.36)	Symptoms	129	3.0
12 up to 18 years														
N children		153			195			242			3030	
N admissions		94			199			239			1579	
N children w admission		58			117			116			1020	
Principal diagnosis	*N*	*%*	*RR (95%CI)*		*N*	*%*	*RR (95%CI)*		*N*	*%*	*RR (95%CI)*		*N*	*%*
Digestive	18	11.8	0.96 (0.61, 1.49)	Digestive	60	30.8	2.50 (1.98, 3.15)	Digestive	60	24.8	2.01 (1.59, 2.56)	Digestive	373	12.3
Injury	18	11.8	1.36 (0.87, 2.12)	Ear	33	16.9	39.44 (21.11, 73.71)	Ear	32	13.2	30.82 (16.39, 57.94)	Injury	263	8.7
Symptoms	§			Injury	23	11.8	1.36 (0.91, 2.03)	Injury	13	5.4	0.62 (0.36, 1.06)	Respiratory	131	4.4
Skin	§			Skin	12	6.2	2.78 (1.53, 5.06)	Symptoms	12	5.0	2.00 (1.10, 3.63)	Symptoms	75	2.5
Musculo-skeletal	§			Factors	11	5.6	4.38 (2.28, 8.42)	Factors	§			Pregnancy	70	2.3
												Musculo-skeletal	70	2.3
18 up to 25 years														
N adults		74			116			121			1609	
N admissions		68			123			138			1642	
N adults w admission		32			57			65			799	

CLO				CLAP				CPO				No OFC		
Principal diagnosis	*N*	*%*	*RR (95%CI)*		*N*	*%*	*RR (95%CI)*		*N*	*%*	*RR (95%CI)*		*N*	*%*
Digestive	18	24.3	1.26 (0.83, 1.90)	Digestive	28	24.1	1.25 (0.89, 1.75)	Digestive	27	22.3	1.15 (0.82, 1.63)	Digestive	311	19.3
Injury	§			Injury	§			Pregnancy	14	11.6	0.90 (0.54, 1.50)	Pregnancy	207	12.9
Pregnancy	§			Pregnancy	§			Injury	12	9.9	0.77 (0.45, 1.35)	Injury	206	12.8
Factors	§			Factors	§			Factors	§			Musculoskeletal	77	4.8
				Respiratory	§			Respiratory	§			Genitourinary	68	4.2

† Diagnoses are coded to ICD-9-CM chapter headings. Most diagnostic groups relate to diseases of body systems (such as Digestive, Respiratory, Musculoskeletal, Genitourinary); Perinatal diagnoses include conditions originating in the perinatal period; Pregnancy diagnoses comprise complications of pregnancy, childbirth and the puerperium; Factors are factors influencing health status and contact with health services, and include such things as specific procedures and after care following treatment for a condition now not present, having a condition that influences health status, personal history of certain diseases; diagnoses categorised as Symptoms include symptoms of various body systems, signs and ill-defined conditions, and non-specific abnormal findings. We included the five most frequent principal diagnoses for each cleft type, listed in descending order of frequency. RR was calculated by comparing the rate for each of CLO, CLAP and CPO, with the rate for the same diagnosis in the group with no OFC, even if that diagnosis was not among the top five for children without OFC.

‡ Denominator includes children with complete follow up over the whole period. § ≤ 10 children admitted, data suppressed. CA, congenital anomaly (other CA excludes orofacial clefts); CI, confidence interval; RR, relative rate of admission compared with rate in the group with no OFC for same diagnosis.