

Trends of ankyloglossia and lingual frenotomy in hospital settings among children in Denmark

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ABSTRACT

INTRODUCTION: It has been proposed that ankyloglossia and lingual frenotomy have increased. The aim of this study was to analyse the trends of ankyloglossia and lingual frenotomy among children treated in hospitals in Denmark in the 1996-2015 period.

METHODS: We included children aged 0-17 years registered with the diagnosis ankyloglossia (DQ381) or the procedure frenotomy (KEJC20) using nationwide hospital-based data from the Danish National Patient Registry in 1996-2015. The incidence during the study period and the average annual percentage change (AAPC) and age-specific incidences were calculated.

RESULTS: A total of 1,608 children were diagnosed with ankyloglossia (67% boys). The overall incidence of ankyloglossia increased from 3.2 per 100,000 in 1996 to 13.6 per 100,000 in 2015, corresponding to an AAPC of 7.1%. The age-specific incidence revealed a more than ten times higher incidence of ankyloglossia among patients less than one year old compared with those aged one year or above. In all, 3,625 frenotomy procedures were carried out (66% boys). The frequency of frenotomy increased from 5.1 per 100,000 in 1996 to 38.5 per 100,000 in 2015, corresponding to an AAPC of 11.1%. The age-specific incidence of frenotomy showed a more than thirty times higher frequency among children less than one year of age.

CONCLUSIONS: Ankyloglossia and frenotomy increased significantly during the period 1996-2015 among children in Denmark according to nationwide hospital data. Rates of both ankyloglossia and frenotomy were highest in children less than one year of age and among boys.

FUNDING: none.

TRIAL REGISTRATION: not relevant.

Diagnosis and management of tongue-tie (ankyloglossia) lack consensus and are controversial [1]. The rates of diagnosis of tongue-tie and of lingual frenotomy procedures have been reported to be increasing in studies from the US and Canada [2, 3]. The prevalence of ankyloglossia among children is estimated to 0.1-12.1% in the US [4]. However, the prevalence of children with ankyloglossia and rates of frenotomy remain unknown among children in Denmark.

Frenotomy is the main surgical procedure used in the treatment of tongue-tie [1]. Frenotomy is often performed in infants with tongue-tie and breastfeeding

problems, although the evidence is low that frenotomy improves breastfeeding [1, 5].

Using nationwide hospital-based data, the aim of this study was to evaluate the national trends in children aged 0-17 years diagnosed with ankyloglossia and undergoing frenotomy procedures in Denmark from 1996 through 2015.

METHODS

Children aged 0-17 years registered with the diagnosis ankyloglossia or the procedure frenotomy in the Danish National Patient Registry (NPR) in the 1996-2015 period were included. The NPR provides information about every contact a patient has at hospitals in Denmark including both in- and out-patient care. The information includes diagnosis code at discharge and the codes of procedures performed during admission, among others. A diagnosis of ankyloglossia was defined as an NPR registration with the International Classification of Diseases, tenth edition (ICD10) code DQ381, and the procedure frenotomy as registration with the SKS code KEJC20 (SKS is a procedure classification system used in the Danish healthcare system).

If the same patient was registered more than once, only the earliest registration of ankyloglossia or frenotomy was included.

Statistical analysis

Statistical analyses were performed in R statistics version 3.4.1 (Stanford University, Stanford, CA, USA) [6]. The incidence rates were calculated as crude incidence rates per 100,000 using the Danish population between 0-17 years of age as reference. Age-specific incidences were calculated using the average population between 1996 and 2015 for the given age as reference. The annual percentage change (APC) and average annual percentage change (AAPC) were calculated using joinpoint trend analysis software v. 4.2.0.2, with growth assumed to be logarithmic with the formula $\ln(y) = xb$. The joinpoint regression analysis estimates possible join points (trend breaks), which are significant changes in trends. APC is the percentage change per year within a given trend, while AAPC is the average percentage change per year for the whole study period. $p \leq 0.05$ were considered statistically significant.

ORIGINAL ARTICLE

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Dan Med J
2020;67(5):A01200051

Trial registration: not relevant.

RESULTS

We identified 1,608 children with the diagnosis ankyloglossia, 66.9% were boys (n = 1,076). The median age at the time of diagnosis was 17 days (interquartile range (IQR): 1 day-3.2 years). The overall incidence of ankyloglossia increased from 3.2 per 100,000 in 1996 to 13.6 per 100,000 in 2015, corresponding to an AAPC of 7.1%. The increase in incidence was non-significantly higher among girls (AAPC = 8.3%) than among boys (AAPC = 6.7%) (Figure 1, Table 1).

Overall and for boys, a non-significant increase was seen in the incidence of ankyloglossia until 2010, which was followed by a significant high increase during 2010 to 2015 (Table 1).

The highest incidence of ankyloglossia was observed among children below one year of age, which had a more than ten times higher incidence of ankyloglossia than older children. Furthermore, we observed a second and third peak among patients at four and 14 years of age (Figure 2).

We identified 3,625 children undergoing frenotomy at hospitals in Denmark during the study period; 65.9% were boys (n = 2,390). The median age when frenotomy was performed was three days (IQR: 1-35 days). The incidence of the frenotomy procedure increased from 5.1 per 100,000 in 1996 to 38.5 per 100,000 in

2015, corresponding to an AAPC of 11.1%. Similar to the incidence of ankyloglossia, the increase in incidence of frenotomy was non-significantly higher among girls (AAPC: 12.9%) than among boys (AAPC 9.8%) (Figure 1, Table 1).

In the trend analysis allowing two trends, a non-significant increase was observed in overall incidence until 2002, followed by a significant, high increase for the remaining part of the study period. A similar pattern was seen for boys and girls separately (Table 1).

The incidence of frenotomy was highest among children less than one year of age, with an incidence that was more than thirty times higher than among the remaining children. A second and third peak were observed at four and 14 years of age (Figure 2).

DISCUSSION

It is uncertain whether ankyloglossia is a congenital oral anomaly requiring treatment or a normal variant [1]. Despite this, we identified an increase in the incidence of children registered with ankyloglossia and more children underwent frenotomy in Danish hospitals. From 1996 to 2015, the incidence of children diagnosed with ankyloglossia more than quadrupled, whereas the incidence of children undergoing the frenotomy procedure increased more than sevenfold. A recent study in the United States showed the same trend with an 834 percent increase in children diagnosed with ankyloglossia and an

FIGURE 1 / Incidence per 100,000 of ankyloglossia (A) and frequency per 100,000 of frenotomy (B) among children aged 0-17 years in Denmark in the 1996-2015 period.

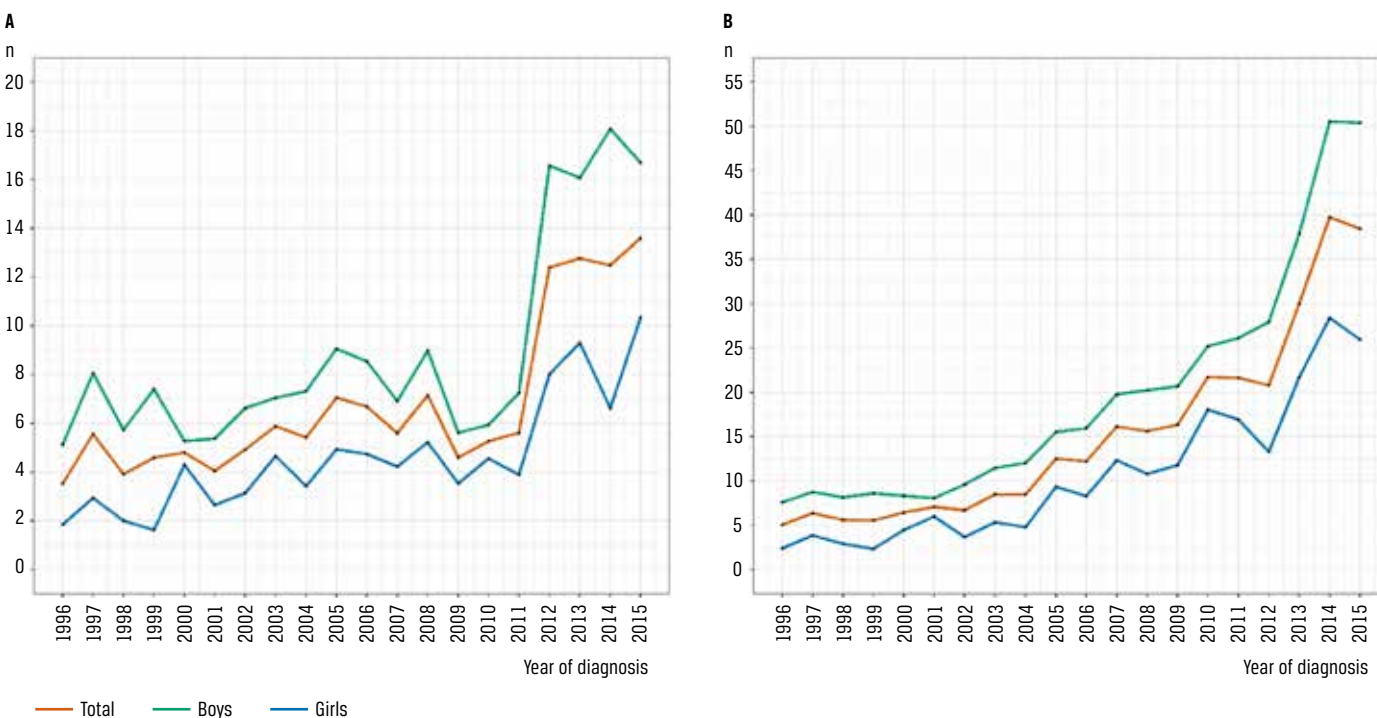


TABLE 1 / Joinpoint trend analysis demonstrating trends in the incidence of ankyloglossia and frequency of frenotomy.

Group	Trend 1			Trend 2			Average	
	period	annual change, % (95%CI)	p-value	period	annual change, % (95%CI)	p-value	annual change, % (95%CI)	p-value
<i>Ankyloglossia</i>								
Total	1996-2010	2.8 (-0.1-5.8)	0.1	2010-2015	19.9 (4.8-37.3)	< 0.05	7.1 (3.1-11.2)	< 0.05
Boys	1996-2010	1.5 (-1.6-4.6)	0.3	2010-2015	22.8 (6.1-42.1)	< 0.05	6.7 (2.4-11.2)	< 0.05
Girls	1996-2011	6.2 (2.8-9.7)	< 0.05	2011-2015	16.8 (-8.6-49.3)	0.2	8.3 (2.7-14.2)	< 0.05
<i>Frenotomy</i>								
Total	1996-2002	5.4 (-0.4-11.6)	0.1	2002-2015	13.8 (11.8-15.8)	< 0.05	11.1 (8.9-13.3)	< 0.05
Boys	1996-2001	1.2 (-4.8-7.4)	0.7	2001-2015	13.1 (11.7-14.6)	< 0.05	9.8 (8.0-11.7)	< 0.05
Girls	1996-1999	4.8 (-24.7-45.9)	0.8	1999-2015	14.5 (11.6-17.4)	< 0.05	12.9 (7.2-18.9)	< 0.05

CI = confidence interval.

866 percent increase in reported frenotomy procedures in children aged 0-17 years from 1997 to 2012 [2].

We observed more boys diagnosed with ankyloglossia, accounting for two thirds of the cases; a distribution that has been reported previously [2, 4, 7]. In the age-specific incidence of ankyloglossia and frequency of frenulotomy, we observed three peaks: One at age zero, one at four years and one at fourteen years (Figure 2). The significant, high incidence at age zero can be explained by ankyloglossia diagnosed and treated with frenotomy in association with breastfeeding difficulties in infants.

The observed increase in ankyloglossia could be related to the global efforts to support breastfeeding; for example, the WHO now recommends breastfeeding as the primary nutrition for infants aged from zero to six months [8]. As 30-40% of new Danish mothers experience breastfeeding difficulties, an increasing attention to the role of ankyloglossia in successful breastfeeding seems probable [9]. However, breastfeeding rates have not changed in Denmark recently, and 97-99.5% of Danish mothers have started breastfeeding right after birth since the late 1980s [10].

We observed a significant increase in children diagnosed with ankyloglossia during the 2010-2015 period (Figure 1). The change in the incidence of ankyloglossia is unlikely to be related to the anatomy of Danish infants or to Danish breastfeeding rates. However, it may possibly be attributed to an increased public awareness. Increased attention in the press and social media has caused more parents to turn to healthcare professionals and alternative therapists deeming tongue tie as the explanation of, e.g., breast feeding problems [11]. The observed increases in both ankyloglossia and frenotomy (Figure 1) might be explained by a shift from diagnosis and treatment in private clinics to a hospital-based service.

Using national hospital registries introduces observer bias. Only children registered at hospital-based

services were included, leaving information about undiagnosed children and those cared for in primary healthcare. The observed bias probably produces an underestimation of both the incidence of ankyloglossia and the frequency of frenotomy in children. However, the reported hospital-based trends in the present study might still be representative for the general trend.

The study may also have registration bias as the diagnosis and procedure codes may be used differently among healthcare professionals. Many more children were undergoing the frenotomy procedure than were registered with a diagnosis of ankyloglossia (Figure 2). This difference may possibly occur because not all infants who are clinically diagnosed with ankyloglossia are correctly registered in the NPR, including the ankyloglossia diagnosis at discharge, even though a frenotomy is performed.

CONCLUSIONS

The incidence of children who were diagnosed with ankyloglossia and underwent frenotomy increased significantly in Denmark in the 1996-2015 period. Boys were more likely to have ankyloglossia and the rates of ankyloglossia were especially high in infants below one year of age. Recent sudden increases may suggest an increased public awareness rather than biological or breastfeeding-related causes.

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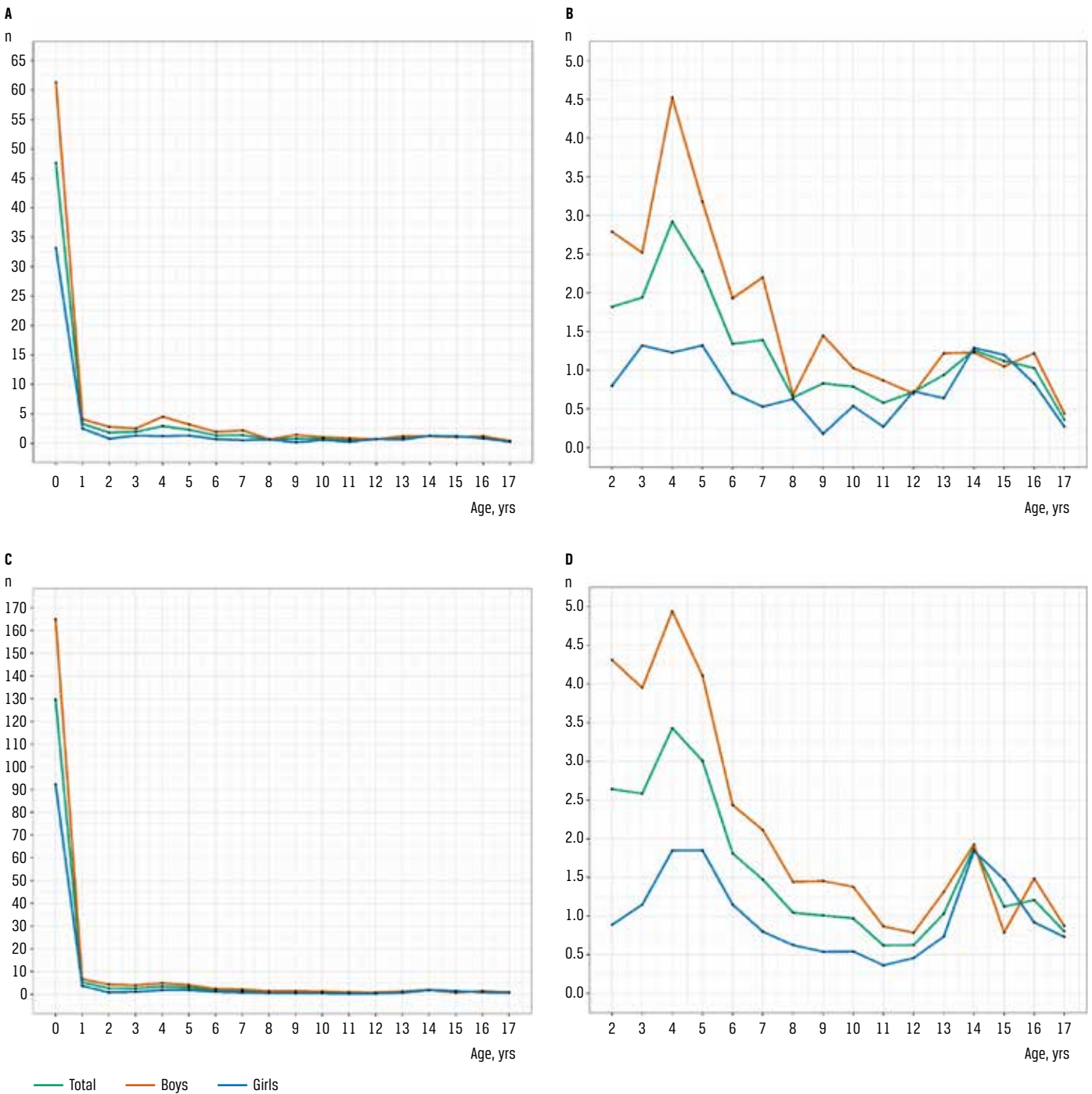
ACCEPTED: 31 March 2020

CONFLICTS OF INTEREST: Disclosure forms provided by the authors are available with the full text of this article at Ugeskriftet.dk/dmj

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FIGURE 2 / Age-specific incidence per 100,000 of ankyloglossia (A, B) and frequency per 100,000 of frenotomy (C, D) in Denmark in the 1996-2015 period.



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