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Complete List of Authors:	Borgström, Anna; Karolinska University Hospital, Huddinge; Karolinska Institutet, Department of Clinical Science, Intervention and Technology, CLINTEC Nerfeldt, Pia; Karolinska University Hospital, Huddinge, ORL department Friberg, Danielle; Karolinska University Hospital, Huddinge, ORL Department Sunnergren, Ola; Department of Otorhinolaryngology, Ryhov County Hospital Stalfors, Joacim; Institute of Clinical Sciences, Sahlgrenska Academy at the University of Gothenburg
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TRENDS AND CHANGES IN THE CLINICAL PRACTICE OF PEDIATRIC TONSIL SURGERY IN SWEDEN 1987 – 2013

Corresponding author: **Anna Borgström**

Postal address: Department of Otorhinolaryngology B53 , Karolinska University Hospital, 141 86 Stockholm, Sweden

Institution: Department of Clinical Science, Intervention and Technology, CLINTEC, Karolinska Institute, Stockholm Sweden

E-mail: anna.borgstrom@karolinska.se

Telephone number: +46-704-220211

Co-authors:

Pia Nerfeldt

Department of Otorhinolaryngology, Karolinska University Hospital

CLINTEC, Karolinska Institute, Stockholm Sweden

Danielle Friberg

Department of Otorhinolaryngology, Karolinska University Hospital

CLINTEC, Karolinska Institute, Stockholm Sweden

Ola Sunnergren

Department of Otorhinolaryngology, Ryhov County Hospital, Jönköping Sweden

Joacim Stalfors

Institute of Clinical Sciences, Sahlgrenska Academy at the University of Gothenburg, Sweden

Key words: Tonsillectomy, Tonsillotomy, Tonsil surgery, Pediatric, Register

Trends and changes in the clinical practice of pediatric tonsil surgery in Sweden: 1987- 2013

Borgström A, Nerfeldt P, Friberg D, Sunnergren O, Stalfors J

Abstract

Objectives: The objective of this study was to longitudinally describe the history of tonsil surgery in Swedish children and adolescents regarding incidence, indications for surgery, surgical methods, and the age- and gender distributions.

Setting: A retrospective longitudinal population-based cohort study based on register data from the Swedish National Patient Register (NPR) and population data from Statistics Sweden.

Participants: All Swedish children 1-<18 years registered in the NPR with a tonsil surgery procedure 1987-2013.

Results: 167,894 tonsil surgeries were registered in the NPR 1987-2013. An increase in the total incidence rate was observed, from 22/10,000 person years in 1987 to 47/10,000 in 2013. The most marked increase was noted in children 1-3 years of age, increasing from 17 to 73/10,000 person years over the period. The proportion children with obstructive/Sleep Disordered Breathing (SDB) indications increased from 42.4% in 1987 to 73.6% in 2013. Partial tonsillectomy, tonsillotomy (TT), increased since 1996 and in 2013 55.1% of all tonsil procedures were TTs.

Conclusions: There have been considerable changes in clinical practice for tonsil surgery in Swedish children over the past decades. Overall, a doubling in the total incidence rate was observed. This increase consisted mainly of an increase in surgical procedures due to obstructive/SDB indications, particularly among the youngest age group (1-3 years old). TT has gradually replaced tonsillectomy as the predominant method for tonsil surgery.

Strengths and limitations of this study

- The major strengths of the study are the large population size and the use of the Swedish National Patient Register (NPR) with its nationwide data observed over a long period of time.
- One limitation is the potential inherent weakness of the NPR with the possibilities of missing values and inaccurate data reporting.

Introduction

Tonsil surgery has an ancient history and was first described in the literature almost 3000 years ago[1]. Today, tonsillectomy (TE), with or without adenoidectomy, is one of the most frequently performed surgical procedures in children, with over 530,000 TEs performed on children in the United States each year [2] and nearly 9000 tonsil procedures performed on children in Sweden in 2013 [3].

Over the past hundred years, tonsil surgery has undergone a number of changes, particularly with respect to indications and surgical methods. Prior to 1900, the predominant surgical technique was tonsillotomy (TT) with subtotal intracapsular removal of the tonsils [4]. Then, along with advances in anaesthesiology at the beginning of the 20th century, total extracapsular TE gradually became more common. This paradigm shift from TT to TE was essentially complete by 1950 [4], and during the second half of the 20th century, TE was the prevailing method. Later, in the 1990s, the previously abandoned method of TT was revived in some countries, including Sweden and Austria [5]. Compared

with TE, TT is associated with fewer perioperative morbidities, such as postoperative haemorrhaging and pain [6 7], and it is now primarily used to treat sleep disordered breathing (SDB) caused by upper airway obstruction due to tonsil hypertrophy [8].
Along with shifts in surgical methods, the indications for tonsil surgery have also changed considerably. At the beginning of the 20th century, in the pre-antibiotic era, infection or recurrent tonsillitis was the main indication for tonsil surgery [9]. Following the introduction of antibiotics in the 1950s, the number of TEs decreased dramatically over the following decades [10 11]. Then, in the 1970s the first cases of pediatric obstructive sleep apnoea (OSA) were identified [12] and a successive shift in indications followed. Today, upper airway obstruction/SDB is the most common indication for tonsil surgery in children, having surpassed the second largest indication group, infectious related tonsil disease [9].
A more detailed analysis of the trends and changes in pediatric tonsil surgery during the 20th century is difficult to perform due to lack of high quality data sources.
The National Board of Health and Welfare in Sweden has initiated a national registry, known as the Swedish National Patient Register (NPR), with the aim of collecting data on all healthcare procedures performed in Sweden [13]. The NPR provides the possibility of performing detailed analyses of trends and changes in health care practices in a nationwide population. Indeed, this database provides a unique source of data with few, if any, equals around the world.
The importance of monitoring historic and current trends in medical practice cannot be overstated. For example, some of the most important changes in indications and methods for pediatric tonsil surgery over the last century were not the result of evidence-based research, related consequences regarding outcome, complications and economic costs are often unstudied.
The objective of this study was to longitudinally describe the history of tonsil surgery in Swedish children and adolescents with respect to incidence, indications for surgery, and surgical methods, as well as age- and gender distributions. A description of the trends in pediatric tonsil surgery is a prerequisite to identifying critical research areas that will allow us to determine best practices and improve health care quality.

Materials and methods

Study design
A retrospective longitudinal population-based cohort study based on register data from the NPR and population data from Statistics Sweden.

Data sources
The Swedish National Patient Register (NPR): All medical data were collected from the NPR. Registration in the NPR is mandatory by law for both public and private care providers (except primary care) in Sweden. The NPR contains information regarding medical data (diagnoses, surgical procedures), patient-related data (gender, age, personal identity number) and information regarding health care providers. The NPR is thought to have complete national coverage for inpatient care starting in 1987 [14]; out-patient care was included beginning in 1997 and was mandatory starting in 2001.
Statistics Sweden: a government agency that coordinates the official statistics of Sweden and provides statistical information on the Swedish population [15].

Study population

The study population included all patients 1-<18 years registered in the NPR with a tonsil surgery procedure between January 1st 1987 and December 31st 2013. The search was based on surgical codes in the Nordic Medico-Statistical Committees Classification of Surgical Procedures, including both TE and TT with or without simultaneous adenoidectomy.

Diagnosis codes from the International Classification of Diseases (ICD) were collected from the NPR. Since 1987, two different ICD-classifications have been used (ICD-9 1987-1996 and ICD-10 1997-). For patients registered from 1997 (ICD-10) onward, it was possible to exclude patients with malignant disease (C- or D 0-48 codes).

Population statistics for all individuals in Sweden aged 1-<18 years during the study period were collected from the Sweden Statistics population database.

Indications for surgery were categorized in two main groups: "Obstructive/SDB" and "Infectious" (**table 1**); all other indications were referred to as "Other." The indication groups were analysed with respect to age, gender and longitudinal incidence.

OBSTRUCTIVE/SDB	INFECTIOUS
ICD-9 (1987-1996)	**ICD-9 (1987-1996)**
307E Specific Sleep Disturbances	101X Vincent´s angina
474B Hypertrophy of adenoids and tonsils	469X Acute nasopharyngitis
474C Adenoid vegetations	462X Acute pharyngitis
519W Other specified diseases of the respiratory system not elsewhere classified	463X Acute tonsillitis
780F Sleep disturbances	465A Acute laryngopharyngitis
786A Dyspnea and respiratory abnormalities	465X Acute upper respiratory infection of unspecified site
786B Stridor	472 Chronic pharyngitis and nasopharyngitis
474B Hypertrophy of adenoids and tonsils	472B Chronic pharyngitis
474C Adenoid vegetations	472C Chronic nasopharyngitis
519W Other specified diseases of the respiratory system not elsewhere classified	474A Chronic tonsillitis
780F Sleep disturbances	475X Peritonsillar abscess
786A Dyspnea and respiratory abnormalities	682B Cellulitis/abscess, neck
786B Stridor	101X Vincent´s angina
474B Hypertrophy of adenoids and tonsils	
ICD-10 (1997-)	**ICD-10 (1997-)**
G47.0 Insomnia	A42.2 Cervicofacial actinomycosis
G47.2 Disorders of the sleep-wake schedule	A42.8 Other forms of actinomycosis
G47.3 Sleep apnoea	A42.9 Actinomycosis, unspecified
G47.30 Sleep apnoea, unspecified	A69.1 Other Vincent infections
G47.39 Sleep apnoea, other	B27.0 Mononucleosis due to Epstein Barr virus
G47.8 Other sleep disorders	B27.1 Cytomegaloviral mononucleosis
G47.9 Sleep disorder, unspecified	B27.8 Other infectious mononucleosis
J35.1 Hypertrophy of tonsils	B27.9 Infectious mononucleosis, unspecified
J35.2 Hypertrophy of adenoids	J00.9 Acute nasopharyngitis
J35.3 Hypertrophy of tonsils with hypertrophy of adenoids	J02.0 Streptococcal pharyngitis
R06.1 Stridor	J02- Acute pharyngitis
R06.3 Periodic breathing	J02.9 Acute pharyngitis, unspecified
R06.5 Mouth breathing	J03.0 Streptococcal tonsillitis
R06.50 Mouth breathing and snoring	J03.8 Acute tonsillitis due to other specified organisms
R06.8 Other and unspecified abnormalities of breathing	J03.9 Acute tonsillitis, unspecified
	J06.9 Acute upper respiratory infection, unspecified
	J31.1 Chronic nasopharyngitis
	J31.2 Chronic pharyngitis
	J35.0 Chronic tonsillitis
	J35.00 Chronic tonsillitis
	J35.09 Chronic tonsillitis

Table 1. *Patients were categorized into "Obstructive/SDB" and "Infectious" groups based on the diagnoses from the ICD-9 and -10 classification.*

Statistical analysis

Only descriptive statistics were used. The denominator used for incidence rate calculations was the sum of the end-of-year population estimates for each year and age.

Results

In total, 167,894 tonsil surgeries (82,398 (49%) girls and 85,496 (51%) boys), were registered in the NPR between 1987 and 2013 (**table 2**).

	1987-1990		1991-1994		1995-1998		1999-2002		2003-2006		2007-2010		2011-2013		1987-2013
Age group	**n**	**rate**	**n**	**rate**	**n**	**rate**	**n**	**Rate**	**n**	**rate**	**n**	**rate**	**n**	**rate**	**n**
F1-3	1 003	16	2 236	31	2 073	33	1 475	28	1 834	32	2 660	42	2 838	56	
F 4-6	2 054	36	3 938	60	4 443	62	2 801	47	2 959	54	3 881	64	3 800	78	
F 7-9	1 375	24	1 937	33	2 341	34	1 671	24	1 353	24	1 700	30	1 568	33	
F 10-12	1 146	20	1 448	25	1 497	24	1 208	17	1 153	17	1 108	19	1 032	23	
F 13-15	1 682	27	1 702	29	1 639	28	1 543	24	1 771	24	1 876	28	1 090	25	
F 16-17	1 687	38	1 621	40	1 481	38	1 535	38	2 153	46	2 592	52	1 494	47	
F subtotal	**8 947**	**26**	**12 882**	**37**	**13 474**	**37**	**10 233**	**28**	**11 223**	**31**	**13 817**	**39**	**11 822**	**45**	**82 398**
M 1-3	1 677	26	3 509	47	3 235	49	2 416	43	2 861	47	4 140	62	4 370	82	
M 4-6	2 934	49	5 278	76	5 979	79	3 793	61	4 017	70	5 549	87	5 051	97	
M 7-9	1 503	25	2 117	34	2 447	34	1 724	23	1 496	25	1 808	30	1 693	34	
M 10-12	790	13	1 081	18	1 008	15	847	11	851	12	863	14	805	17	
M 13-15	746	11	898	15	866	14	730	11	805	10	1 006	15	630	14	
M 16-17	766	17	769	18	672	16	768	18	1 038	21	1 235	24	725	21	
M subtotal	**8 416**	**24**	**13 652**	**37**	**14 207**	**37**	**10 278**	**27**	**11 068**	**29**	**14 601**	**39**	**13 274**	**47**	**85 496**
Total	**17 363**	**25**	**26 534**	**37**	**27 681**	**37**	**20 511**	**28**	**22 291**	**30**	**28 418**	**39**	**25 096**	**46**	**167 894**

Table 2. *The number and incidence rate of tonsil surgery procedures registered in the NPR between 1987 and 2013 by age, gender (F=females, M=males) and time period. The rate was calculated as the incidence/10,000 person years.*

Incidence

The overall incidence rate of registered tonsil operations in patients aged 1-<18 years increased from 22/10,000 person years in 1987 to 47/10,000 person years in 2013 (**figure 1**). This increase was continuous over the study period, with the exception of 1996 through 2001, when a decrease was observed (**figure 1**).

The most marked increase in incidence was noted in children aged 1-3 years, increasing from 17/10,000 person years in 1987 to 73/10,000 person years in 2013. This 1-3 year-old group increased their share of total tonsil surgeries from 13.5% 1987 to 29.4% in 2013.

Gender and age

Figures 2a and b show the age and gender distributions as well as the indication groups. For both genders, the indication "obstructive/SDB" was most common in the younger age groups, with a peak between ages 2 to 6, and the highest incidence (113/10,000 person years) was in 3-year-old boys with obstruction between 2008 and 2013 (***figure 2 a, b***). Infectious indications were more common in teenagers for both genders, particularly for girls, whose incidence was 3-4 times greater compared with boys of the same age. Further, **figures 2a and b** show that the highest incidence levels for tonsil surgery for boys and girls at all ages were observed over the last time period, 2008-2013. The major reason for this increase was operations in young children with obstruction.

Indications

The indications for surgery between 1987 and 2013 are presented as proportions of the total incidence in **figure 1**. The incidence of tonsil procedures performed for the indication obstructive/SDB increased nearly four-fold over this period, from 10 to 36/10,000 person years (**figure 1)**. Since 1990, obstruction/SDB constituted the major indication group. In 1987, 42.4% of tonsil procedures were performed for obstructive/SDB, whereas in 2013, this proportion was 73.6%. In the youngest age groups (children 1-<4 years old), the vast majority (>90%) of tonsil procedures were performed for obstructive/SDB indications. The incidence of infectious tonsil disease as an indication remained stable over the study period.

Surgical methods

Figure 3 shows the proportions of TE and TT (+/- adenoidectomy) between 1987 and 2013. An increase in the number of TT procedures began in 1996 with a gradual annual increasing trend, followed by a sharp increase from 2006 onward. From 1997 to 2005, TT increased from 1.8% to 9.8% of all tonsil procedures, and from 2006 to 2013 it increased from 11.2% to 55.1% of all procedures. From 2011 onwards, TT was more common than TE. The major indication for TT was obstruction, and in 2013 >96% of all TTs were performed for this indication. When TE was utilized, about half (49%) of the procedures were performed due to infection in 2013, a percentage that was generally stable during the observed period.

In the youngest group (1-3 year olds), TT was most common, and in 2013, 71% of all tonsil surgeries in this age group were TTs.

Discussion

This study was based on a national cohort and describes the epidemiological trends during 1987-2013 for tonsil surgeries in Swedish children. Overall, the incidence rate for pediatric tonsil surgery has roughly doubled during the observation period due to an increase in the number of procedures performed for the obstruction/SDB indication. Furthermore, TT has gradually replaced TE as the predominant surgical method. Few previous studies have addressed these topics in large national cohorts over long periods of time.

Incidence

During the study period, 1987-2013, there was a near doubling of the incidence rate for tonsil operations in Swedish children, with incidence rates reaching 47/10,000 person years in 2013. A similar, albeit weaker, trend was reported in a Danish study based on a

national cohort, where the cumulative risk of tonsillectomy in the first 20 years of life increased nearly 20% between 1980 and 2001 [16].

Another important observation in the present study was that the incidence for the youngest group of children, 1-3 years of age, increased more than four-fold over the study period. This trend was also seen in a study from the UK in which TE rates among children <4 years of age increased from 13.5 to 21.3/10,000 children during the period from 2001 to 2012 [17].

The Swedish incidence rate can be viewed as relatively low compared with other nations. Van den Akker et al. reported large differences in tonsillectomy rates between several European countries, the US, Canada and Australia. Incidence rates varied from 19/10,000 person-years in Canada to 118/10,000 in Northern Ireland [18]. The incidence rates in our study varied from 22/10,000 person-years in 1987 to 47/10,000 in 2013. Potential explanations to this variation in incidence rates between different countries include differences in national guidelines for tonsil surgery, differences in the availability of medical service, and differences in reimbursement systems.

For instance, in the Netherlands, antibiotics are not recommended for uncomplicated tonsillitis, which could be a reason for the relative higher incidence of tonsillectomy in that country [19]. By contrast, in the NHS system of the UK, the incidence rate has decreased after evidence regarding outcomes following tonsil surgery for infectious indications were questioned. Concomitant with this decrease in TE rates, there was an increase in admissions and serious complications due to tonsil infections [20].

Interestingly, local geographic and socio-demographic factors have been reported to influence tonsil surgery incidence rates [17 21]. A recent Swedish study reported an increased risk of pediatric SDB in families with low socio-economic status [22], and this has also been suggested to be a risk factor for group A streptococcal infections [23].

Age and gender

We observed a peak in incidence rates during the preschool years, the same age that the prevalence of SDB/OSA peaks [24]. This peak was higher for boys than it was for girls, which is likely because childhood SDB is more common in boys [25]. Another peak occurred in teenage girls with primarily infectious indications. This difference in gender distributions among teenagers could indicate that infectious tonsil diseases are more prevalent in females, although the reasons for this are not fully understood. Overall the observed age and gender distributions were well in line with previous publications showing similar results [21 26].

Indications

Overall, a shift in the prevalence of indications was observed, with obstructive/SDB indications increasing over the study period. Obstruction/SDB now constitutes the primary indication for tonsil surgery in children, consistent with other published studies [27 28]. The incidence for infectious indications remained relatively stable over the study period.

The most common indication in the younger age group by far was obstruction/SDB, which was seen in this study as well as in several previous studies [10 28], likely reflecting an increased awareness of upper airway obstruction and SDB as a common disease in the pediatric population. In a single county study from Minnesota, a comparable large-scale shift in indications was observed with an increase in the percentage of SDB/obstructive indications from 12% in 1970 to 77% in 2005 [26]. Bhattacharyya et al. reported that the rates of paediatric adenotonsilletomy nearly doubled between 1996 and 2006, likely due to increased recognition of SDB and obstructive sleep apnoea (OSA) [29].

Surgical methods

Along with a shift in indications, there has also been a shift in surgical methods, with TT gradually replacing TE as the most common surgical method for children in Sweden. This was also the conclusion of a study based on data from another Swedish registry independent of the NPR, the National Tonsil Register in Sweden [30]. Since 2011, TT has become more common than TE in Sweden. That same year, new national guidelines for TT were issued in Sweden [31]. One possible explanation for our findings could be that Swedish ENT surgeons consider TT to be safe enough to perform on small children. TT has advantages over TE with respect to postoperative complications such as pain and bleeding [6 32] with comparable outcomes for symptom relief [33]. The clinical practice of TT for treating airway obstructions in children is spreading; for instance, in Austria, several serious complications due to posttonsillectomy haemorrhaging has led to a switch to TT [5].

Strengths and limitations

In this study, there was a marked decrease in the incidence rate of tonsil surgeries between 1996 and 2008. The reasons for this decrease in the number of recorded surgeries were analysed together with the representatives from the NPR [34]. One reason for this dip could be a failure on the part of the NPR to collect data on performed surgeries, perhaps due to the increased use of day-care surgery between 1997 and 2001. Another reason could be insufficient reporting to the NPR from private hospitals, which became more numerous during this period. However, we believe this decrease was too great to be explained by failed registrations alone and must represent an actual decrease in the number of surgeries, although the true extent of this decrease cannot be precisely determined.

The NPR is generally considered to have high coverage [14], although like all clinical registers, it has potential inherent limitations, including inaccurate data reporting and missing values.

Major strengths include the large population size of the register and its wealth of unique nationwide data over a long time period. It can therefore be used to determine actual longitudinal application of a clinical practice in a specific country over long periods of time.

Conclusions

Our results showed considerable changes in clinical practices for tonsil surgery in Sweden during the period from 1987 to 2013. Overall, a doubling in the incidence rates was observed during this period. The reason for this change was an increase in surgical procedures due to upper airway obstruction/SDB, particularly among the youngest age group (1-3 years old) and among boys. Further, TT has gradually replaced TE as the predominant surgical method for obstructive/SDB indications. The incidence rates for infectious indications for tonsil surgery have remained relatively stable during the study period, and the most common patients in the infectious group were teenage girls.

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Footnotes

Ethics: This study was approved by the regional ethical review board of Gothenburg, Sweden (Dnr 534-14).

Contributorship statement: AB and JS contributed to the study design. AB, PN, DF, OS and JS contributed to the interpretation and analysis of data. AB wrote the first draft of the manuscript. PN, DF, OS and JS contributed to the final writing and revising of the manuscript, and checked for important intellectual content. All authors approved of the final manuscript as submitted.

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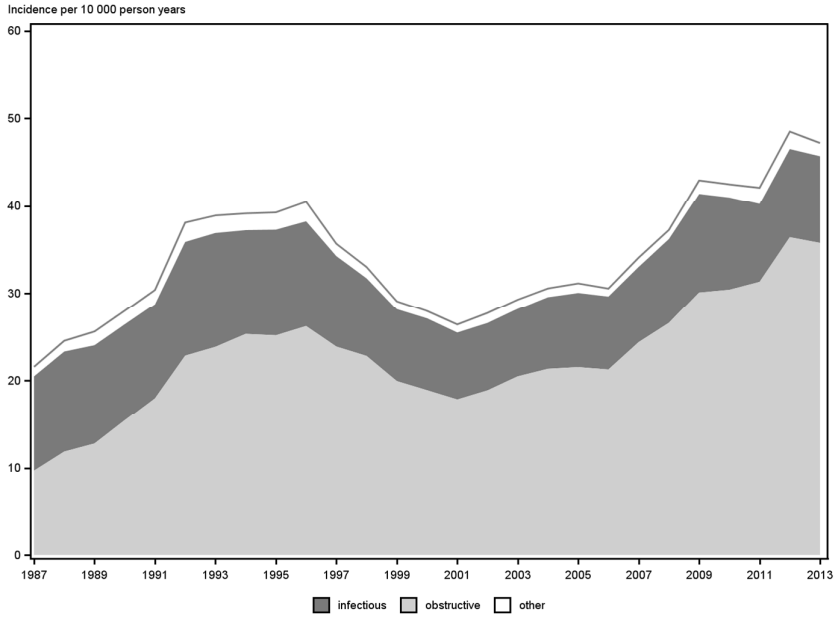
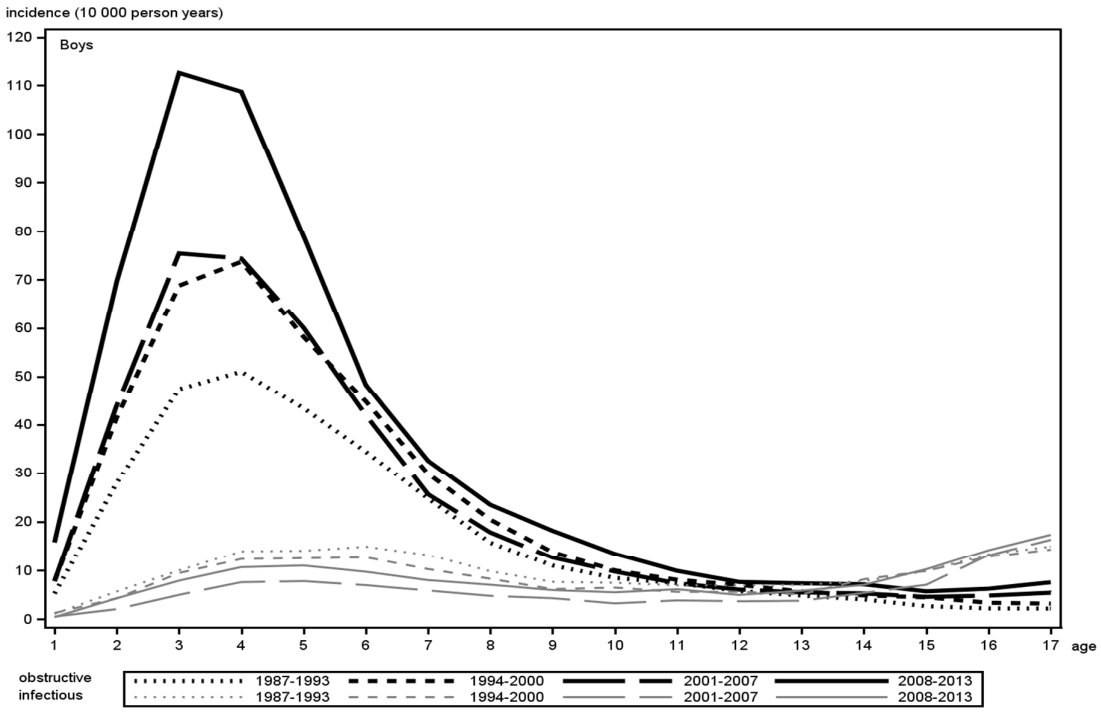




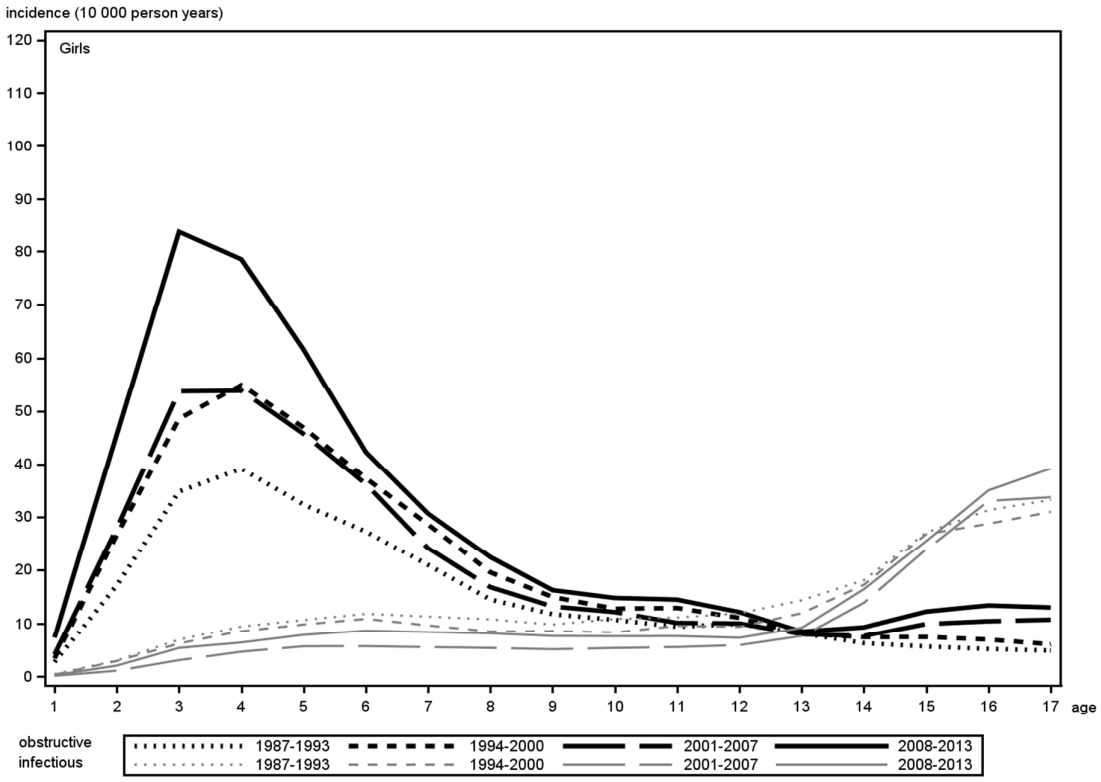
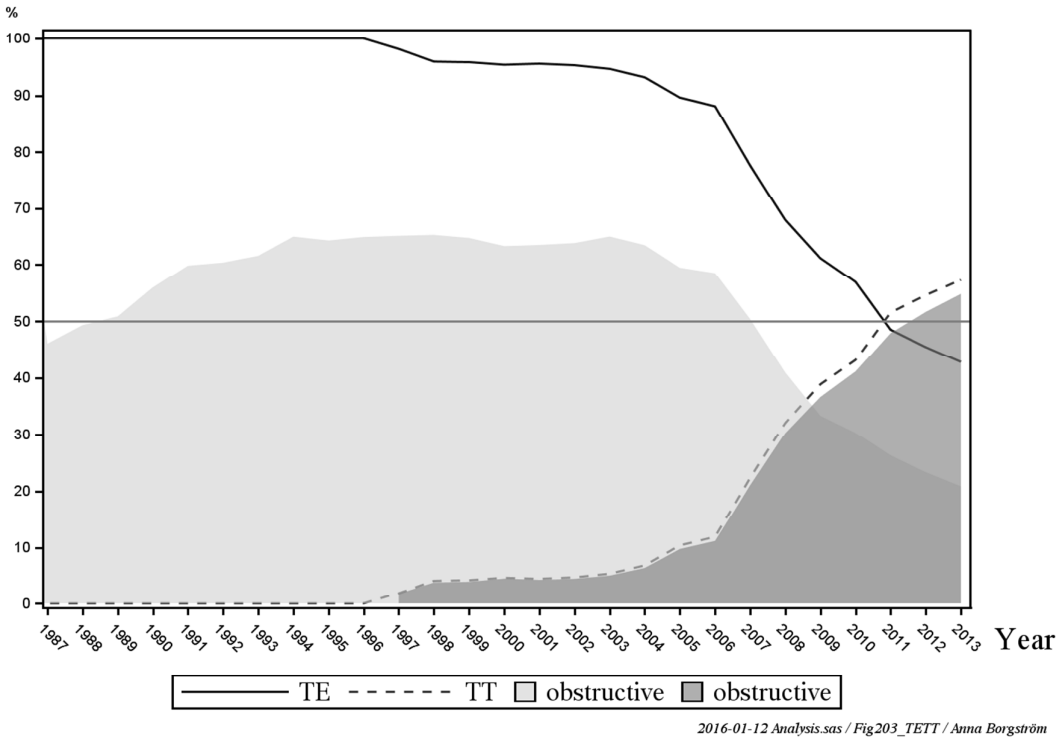


Fig 3. Operation method



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TRENDS AND CHANGES IN PEDIATRIC TONSIL SURGERY IN SWEDEN 1987 – 2013: A POPULATION-BASED COHORT STUDY

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TRENDS AND CHANGES IN PEDIATRIC TONSIL SURGERY IN SWEDEN 1987 – 2013: A POPULATION-BASED COHORT STUDY

Corresponding author: **Anna Borgström**

Postal address: Department of Otorhinolaryngology B53, Karolinska University Hospital, 141 86 Stockholm, Sweden

Institution: Department of Clinical Science, Intervention and Technology, CLINTEC, Karolinska Institute, Stockholm Sweden

E-mail: anna.borgstrom@karolinska.se

Telephone number: +46-704-220211

Co-authors:

Pia Nerfeldt

Department of Otorhinolaryngology, Karolinska University Hospital

CLINTEC, Karolinska Institute, Stockholm Sweden

Danielle Friberg

Department of Otorhinolaryngology, Karolinska University Hospital

CLINTEC, Karolinska Institute, Stockholm Sweden

Ola Sunnergren

Department of Otorhinolaryngology, Ryhov County Hospital, Jönköping Sweden

Joacim Stalfors

Institute of Clinical Sciences, Sahlgrenska Academy at the University of Gothenburg, Sweden

Key words: Tonsillectomy, Tonsillotomy, Tonsil surgery, Pediatric, Register

Trends and changes in pediatric tonsil surgery in Sweden 1987- 2013: a population-based cohort study

Borgström A, Nerfeldt P, Friberg D, Sunnergren O, Stalfors J

Abstract

Objectives: The objective of this study was to longitudinally describe the history of tonsil surgery in Swedish children and adolescents regarding incidence, indications for surgery, surgical methods, and the age- and gender distributions.

Setting: A retrospective longitudinal population-based cohort study based on register data from the Swedish National Patient Register (NPR) and population data from Statistics Sweden.

Participants: All Swedish children 1-<18 years registered in the NPR with a tonsil surgery procedure 1987-2013.

Results: 167,894 tonsil surgeries were registered in the NPR 1987-2013. An increase in the total incidence rate was observed, from 22/10,000 person years in 1987 to 47/10,000 in 2013. The most marked increase was noted in children 1-3 years of age, increasing from 17 to 73/10,000 person years over the period. The proportion children with obstructive/Sleep Disordered Breathing (SDB) indications increased from 42.4% in 1987 to 73.6% in 2013. Partial tonsillectomy, tonsillotomy (TT), increased since 1996 and in 2013 55.1% of all tonsil procedures were TTs.

Conclusions: There have been considerable changes in clinical practice for tonsil surgery in Swedish children over the past decades. Overall, a doubling in the total incidence rate was observed. This increase consisted mainly of an increase in surgical procedures due to obstructive/SDB indications, particularly among the youngest age group (1-3 years old). TT has gradually replaced tonsillectomy as the predominant method for tonsil surgery.

Strengths and limitations of this study

- This study describes the epidemiological trends of tonsil surgery in Swedish children over the past decades.
- The major strengths of the study are the large population size and the use of the Swedish National Patient Register (NPR) with unique nationwide data and high coverage.
- The study covers a long period of time and can therefore be used to determine actual longitudinal application of clinical practice concerning tonsil surgery in Sweden.
- One limitation is the potential inherent weakness of the NPR with the possibilities of missing values and inaccurate data reporting.
- Data are limited to a Swedish population and the generalisability from a global perspective might be somewhat limited, but the results can possibly indicate the evolutionary factors affecting tonsil surgery on a more global scale.

Introduction

Tonsil surgery has an ancient history and was first described in the literature almost 3000 years ago[1]. Today, tonsillectomy (TE), with or without adenoidectomy, is one of the most frequently performed surgical procedures in children, with over 530,000 TEs performed

on children in the United States each year [2] and nearly 9000 tonsil procedures performed on children in Sweden in 2013 [3].

Over the past hundred years, tonsil surgery has undergone a number of changes, particularly with respect to indications and surgical methods. Prior to 1900, the predominant surgical technique was tonsillotomy (TT) with subtotal intracapsular removal of the tonsils [4]. Then, along with advances in anaesthesiology at the beginning of the 20th century, total extracapsular TE gradually became more common. This paradigm shift from TT to TE was essentially complete by 1950 [4], and during the second half of the 20th century, TE was the prevailing method. Later, in the 1990s, the previously abandoned method of TT was revived in some countries, including Sweden and Austria [5]. Compared with TE, TT is associated with fewer perioperative morbidities, such as postoperative haemorrhage and pain [6 7], and it is now primarily used to treat sleep disordered breathing (SDB) caused by upper airway obstruction due to tonsil hypertrophy [8].

Along with shifts in surgical methods, the indications for tonsil surgery have also changed considerably. At the beginning of the 20th century, in the pre-antibiotic era, infection or recurrent tonsillitis was the main indication for tonsil surgery [9]. Following the introduction of antibiotics in the 1950s, the number of TEs decreased dramatically over the following decades [10 11]. Then, in the 1970s the first cases of pediatric obstructive sleep apnoea (OSA) were identified [12] and a successive shift in indications followed. Today, upper airway obstruction/SDB is the most common indication for tonsil surgery in children, having surpassed the second largest indication group, infectious related tonsil disease [9].

A more detailed analysis of the trends and changes in pediatric tonsil surgery during the 20th century is difficult to perform due to lack of high quality data sources.

The National Board of Health and Welfare in Sweden has initiated a national registry, known as the Swedish National Patient Register (NPR), with the aim of collecting data on all healthcare procedures performed in Sweden [13]. The NPR provides the possibility of performing detailed analyses of trends and changes in health care practices in a nationwide population. Indeed, this database provides a unique source of data with few, if any, equals around the world.

The importance of monitoring historic and current trends in medical practice cannot be overstated. Some of the most important changes in indications and methods for pediatric tonsil surgery over the last century were not the result of evidence-based research, rather they were based on local praxis and traditions. Furthermore, the related consequences of these changes regarding outcome, complications and economic costs are often unstudied.

The objective of this study was to longitudinally describe the history of tonsil surgery in Swedish children and adolescents with respect to incidence, indications for surgery, and surgical methods, as well as age- and gender distributions. A description of the trends in pediatric tonsil surgery is a prerequisite to identifying critical research areas that will allow us to determine best practices and improve health care quality and can also be a guide to future public health planning.

Materials and methods

Study design

A retrospective longitudinal population-based cohort study based on register data from the NPR and population data from Statistics Sweden.

Data sources

The Swedish National Patient Register (NPR): All medical data were collected from the NPR. Registration in the NPR is mandatory by law for both public and private care

providers (except primary care) in Sweden. The NPR contains information regarding medical data (diagnoses, surgical procedures), patient-related data (gender, age, personal identity number) and information regarding health care providers. The NPR is thought to have complete national coverage for inpatient care starting in 1987 [14]; out-patient care was included beginning in 1997 and was mandatory starting in 2001.

Statistics Sweden: a government agency that coordinates the official statistics of Sweden and provides statistical information on the Swedish population [15].

Study population

The study population included all patients 1-<18 years registered in the NPR with a tonsil surgery procedure between January 1st 1987 and December 31st 2013. The search was based on surgical codes in the Nordic Medico-Statistical Committees Classification of Surgical Procedures, including both TE and TT with or without simultaneous adenoidectomy.

Diagnosis codes from the International Classification of Diseases (ICD) were collected from the NPR. Since 1987, two different ICD-classifications have been used (ICD-9 1987-1996 and ICD-10 1997-). For patients registered from 1997 (ICD-10) onward, it was possible to exclude patients with malignant disease (C- or D 0-48 codes).

Population statistics for all individuals in Sweden aged 1-<18 years during the study period were collected from the Sweden Statistics population database.

Indications for surgery were categorized in two main groups: “Obstructive/SDB” and “Infectious” (**table 1**); all other indications were referred to as “Other.” The indication groups were analysed with respect to age, gender and longitudinal incidence.

OBSTRUCTIVE/SDB	INFECTIOUS
ICD-9 (1987-1996)	**ICD-9 (1987-1996)**
307E Specific Sleep Disturbances	101X Vincent´s angina
474B Hypertrophy of adenoids and tonsils	469X Acute nasopharyngitis
474C Adenoid vegetations	462X Acute pharyngitis
519W Other specified diseases of the respiratory system not elsewhere classified	463X Acute tonsillitis
780F Sleep disturbances	465A Acute laryngopharyngitis
786A Dyspnea and respiratory abnormalities	465X Acute upper respiratory infection of unspecified site
786B Stridor	472 Chronic pharyngitis and nasopharyngitis
474B Hypertrophy of adenoids and tonsils	472B Chronic pharyngitis
474C Adenoid vegetations	472C Chronic nasopharyngitis
519W Other specified diseases of the respiratory system not elsewhere classified	474A Chronic tonsillitis
780F Sleep disturbances	475X Peritonsillar abscess
786A Dyspnea and respiratory abnormalities	682B Cellulitis/abscess, neck
786B Stridor	101X Vincent´s angina
474B Hypertrophy of adenoids and tonsils	
ICD-10 (1997-)	**ICD-10 (1997-)**
G47.0 Insomnia	A42.2 Cervicofacial actinomycosis
G47.2 Disorders of the sleep-wake schedule	A42.8 Other forms of actinomycosis
G47.3 Sleep apnoea	A42.9 Actinomycosis, unspecified
G47.30 Sleep apnoea, unspecified	A69.1 Other Vincent infections
G47.39 Sleep apnoea, other	B27.0 Mononucleosis due to Epstein Barr virus
G47.8 Other sleep disorders	B27.1 Cytomegaloviral mononucleosis
G47.9 Sleep disorder, unspecified	B27.8 Other infectious mononucleosis
J35.1 Hypertrophy of tonsils	B27.9 Infectious mononucleosis, unspecified
J35.2 Hypertrophy of adenoids	J00.9 Acute nasopharyngitis
J35.3 Hypertrophy of tonsils with hypertrophy of adenoids	J02.0 Streptococcal pharyngitis
R06.1 Stridor	J02- Acute pharyngitis
R06.3 Periodic breathing	J02.9 Acute pharyngitis, unspecified
R06.5 Mouth breathing	J03.0 Streptococcal tonsillitis
R06.50 Mouth breathing and snoring	J03.8 Acute tonsillitis due to other specified organisms
R06.8 Other and unspecified abnormalities of breathing	J03.9 Acute tonsillitis, unspecified
	J06.9 Acute upper respiratory infection, unspecified
	J31.1 Chronic nasopharyngitis
	J31.2 Chronic pharyngitis
	J35.0 Chronic tonsillitis
	J35.00 Chronic tonsillitis
	J35.09 Chronic tonsillitis

Table 1. *Patients were categorized into "Obstructive/SDB" and "Infectious" groups based on the diagnoses from the ICD-9 and -10 classification.*

Statistical analysis

Mainly descriptive statistics were used. The denominator used for incidence rate calculations was the sum of the end-of-year population estimates for each year and age. For comparison of the event rate between boys and girls, assuming that the events are Poisson distributed, an exact binomial test was performed.

Gender differences regarding indication for surgery in specific age groups were tested using the Cochran-Mantel-Haenszel test.

Incidence rate trends were tested by using joinpoint regression models (Joinpoint Regression program, version 4.3.1.0 – April 2016; Statistical Methodology and Applications Branch, Surveillance Research Program, National Cancer Institute), that identified points (joinpoints) where linear trends of tonsil surgery changed significantly in direction or magnitude. The rate of change for each trend was tested to determine whether the change was significantly different from zero. In the final model each trend is described by an annual percentage change (APC) with a 95% confidence interval (CI). The joinpoint analyses were performed for boys and girls separately.
All significance tests were two-sided and conducted at the 5% significance level. All analyses except the Joinpoint regression were performed with SAS System Version 9, SAS Inst., Cary, NC, US.

Results

In total, 167,894 tonsil surgeries (82,398 (49%) girls and 85,496 (51%) boys), were registered in the NPR between 1987 and 2013 (**table 2**).

	1987-1990		1991-1994		1995-1998		1999-2002		2003-2006		2007-2010		2011-2013		1987-2013
Age group	**n**	**rate**	**n**	**rate**	**n**	**rate**	**n**	**Rate**	**n**	**rate**	**n**	**rate**	**n**	**rate**	**n**
F1-3	1 003	16	2 236	31	2 073	33	1 475	28	1 834	32	2 660	42	2 838	56	
F 4-6	2 054	36	3 938	60	4 443	62	2 801	47	2 959	54	3 881	64	3 800	78	
F 7-9	1 375	24	1 937	33	2 341	34	1 671	24	1 353	24	1 700	30	1 568	33	
F 10-12	1 146	20	1 448	25	1 497	24	1 208	17	1 153	17	1 108	19	1 032	23	
F 13-15	1 682	27	1 702	29	1 639	28	1 543	24	1 771	24	1 876	28	1 090	25	
F 16-17	1 687	38	1 621	40	1 481	38	1 535	38	2 153	46	2 592	52	1 494	47	
F subtotal	**8 947**	**26**	**12 882**	**37**	**13 474**	**37**	**10 233**	**28**	**11 223**	**31**	**13 817**	**39**	**11 822**	**45**	**82 398**
M 1-3	1 677	26	3 509	47	3 235	49	2 416	43	2 861	47	4 140	62	4 370	82	
M 4-6	2 934	49	5 278	76	5 979	79	3 793	61	4 017	70	5 549	87	5 051	97	
M 7-9	1 503	25	2 117	34	2 447	34	1 724	23	1 496	25	1 808	30	1 693	34	
M 10-12	790	13	1 081	18	1 008	15	847	11	851	12	863	14	805	17	
M 13-15	746	11	898	15	866	14	730	11	805	10	1 006	15	630	14	
M 16-17	766	17	769	18	672	16	768	18	1 038	21	1 235	24	725	21	
M subtotal	**8 416**	**24**	**13 652**	**37**	**14 207**	**37**	**10 278**	**27**	**11 068**	**29**	**14 601**	**39**	**13 274**	**47**	**85 496**
Total	**17 363**	**25**	**26 534**	**37**	**27 681**	**37**	**20 511**	**28**	**22 291**	**30**	**28 418**	**39**	**25 096**	**46**	**167 894**
p-value															**0.0011**

Table 2*. The number and incidence rate of tonsil surgery procedures registered in the NPR between 1987 and 2013 by age, gender (F=females, M=males) and time period. The rate was calculated as the incidence/10,000 person years. The p-value for the gender difference is shown.*

Incidence

The overall incidence rate of registered tonsil operations in patients aged 1-<18 years increased from 22/10,000 person years in 1987 to 47/10,000 person years in 2013 (**figure 1**). This increase was continuous over the study period, with the exception of 1996 through 2001, when a decrease was observed (**figure 1**).

The most marked increase in incidence was noted in children aged 1-3 years, increasing from 17/10,000 person years in 1987 to 73/10,000 person years in 2013. This 1-3 year-old group increased their share of total tonsil surgeries from 13.5% 1987 to 29.4% in 2013.

The trends in incidence rates for boys and girls are shown in **figure 2a+b**. Trends were similar and significant for both genders (***table 3***).

3a(Boys)

Lower end point	Upper endpoint	APC	Lower 95% CI	Upper 95% CI	p-value
1987	1994	12.0	9.3	14.8	**<0.001**
1994	2001	-7.7	-10.2	-5.1	**<0.001**
2001	2013	6.0	5.0	6.9	**<0.001**

3b(Girls)

Lower end point	Upper endpoint	APC	Lower 95% CI	Upper 95% CI	p-value
1987	1995	7.1	5.3	8.9	**<0.001**
1995	2001	-7.2	-10.2	-4.2	**<0.001**
2001	2013	4.9	4.0	5.8	**>0.001**

Table 3a+b*. Summary of joinpoint annual percentage change (APC) for tonsil surgery incidence in boys (3a) and girls (3b) 1987-2013. The APC is significant different from zero at alpha=0.05. CI, Confidence interval.*

Gender and age

For the total time period the incidence difference between the genders were significant (p=0.0011, exact binomial test), showing a dominance of boys **(table 2)**.

Figures 3a and b show the age and gender distributions as well as the indication groups. For both genders, the indication "obstructive/SDB" was most common in the younger age groups, with a peak between ages 2 to 6, and the highest incidence (113/10,000 person years) was in 3-year-old boys with obstruction between 2008 and 2013 (***figure 3 a, b***). Infectious indications were more common in teenagers for both genders, particularly for girls, whose incidence was 3-4 times greater compared with boys of the same age.

The gender differences were significant in the age group 2-6 year olds who were operated for obstructive/SDB indications with a male dominance (p<0.001) as well as in the age group 14-17 year olds operated for infection with a female dominance (p<0.001, Cochran-Mantel-Haenszel test).

Further, **figures 3a and b** show that the highest incidence levels for tonsil surgery for boys and girls at all ages were observed over the last time period, 2008-2013. The major reason for this increase was operations in young children with obstruction.

Indications

The indications for surgery between 1987 and 2013 are presented as proportions of the total incidence in **figure 1**. The incidence of tonsil procedures performed for the indication obstructive/SDB increased nearly four-fold over this period, from 10 to

36/10,000 person years (**figure 1)**. Since 1990, obstruction/SDB constituted the major indication group. In 1987, 42.4% of tonsil procedures were performed for obstructive/SDB, whereas in 2013, this proportion was 73.6%. In the youngest age groups (children 1-<4 years old), the vast majority (>90%) of tonsil procedures were performed for obstructive/SDB indications. The incidence of infectious tonsil disease as an indication remained stable over the study period.

Surgical methods

Figure 4 shows the proportions of TE and TT (+/- adenoidectomy) between 1987 and 2013. An increase in the number of TT procedures began in 1996 with a gradual annual increasing trend, followed by a sharp increase from 2006 onward. From 1997 to 2005, TT increased from 1.8% to 9.8% of all tonsil procedures, and from 2006 to 2013 it increased from 11.2% to 55.1% of all procedures. From 2011 onwards, TT was more common than TE. The major indication for TT was obstruction, and in 2013 >96% of all TTs were performed for this indication. When TE was utilized, about half (49%) of the procedures were performed due to infection in 2013, a percentage that was generally stable during the observed period.

In the youngest group (1-3 year olds), TT was most common, and in 2013, 71% of all tonsil surgeries in this age group were TTs.

Discussion

This study was based on a national cohort and describes the epidemiological trends during 1987-2013 for tonsil surgeries in Swedish children. Overall, the incidence rate for pediatric tonsil surgery has roughly doubled during the observation period due to an increase in the number of procedures performed for the obstruction/SDB indication. Furthermore, TT has gradually replaced TE as the predominant surgical method. Few previous studies have addressed these topics in large national cohorts over long periods of time.

Incidence

During the study period, 1987-2013, there was a near doubling of the incidence rate for tonsil operations in Swedish children, with incidence rates reaching 47/10,000 person years in 2013. A similar, albeit weaker, trend was reported in a Danish study based on a national cohort, where the cumulative risk of tonsillectomy in the first 20 years of life increased nearly 20% between 1980 and 2001 [16].

Another important observation in the present study was that the incidence for the youngest group of children, 1-3 years of age, increased more than four-fold over the study period. This trend was also seen in a study from the UK in which TE rates among children <4 years of age increased from 13.5 to 21.3/10,000 children during the period from 2001 to 2012 [17].

The Swedish incidence rate can be viewed as relatively low compared with other nations. Van den Akker et al. reported large differences in tonsillectomy rates between several European countries, the US, Canada and Australia. Incidence rates varied from 19/10,000 person-years in Canada to 118/10,000 in Northern Ireland [18]. The incidence rates in our study varied from 22/10,000 person-years in 1987 to 47/10,000 in 2013. Potential explanations to this variation in incidence rates between different countries include differences in national guidelines for tonsil surgery, differences in the availability of medical service, and differences in reimbursement systems.

For instance, in the Netherlands, antibiotics are not recommended for uncomplicated tonsillitis, which could be a reason for the relative higher incidence of tonsillectomy in that country [19]. By contrast, in the NHS system of the UK, the incidence rate has decreased after evidence regarding outcomes following tonsil surgery for infectious indications were questioned. Concomitant with this decrease in TE rates, there was an increase in admissions and serious complications due to tonsil infections [20].
Interestingly, local geographic and socio-demographic factors have been reported to influence tonsil surgery incidence rates [17 21]. A recent Swedish study reported an increased risk of pediatric SDB in families with low socio-economic status [22], and this has also been suggested to be a risk factor for group A streptococcal infections [23].

Age and gender
We observed a peak in incidence rates during the preschool years, the same age that the prevalence of SDB/OSA peaks [24]. This peak was significantly higher for boys than it was for girls, which is likely because childhood SDB is more common in boys [25]. Another peak occurred in teenage girls with primarily infectious indications. This significant difference in gender distributions among teenagers could indicate that infectious tonsil diseases are more prevalent in females, although the reasons for this are not fully understood. Overall the observed age and gender distributions were well in line with previous publications showing similar results [21 26].

Indications
Overall, a shift in the prevalence of indications was observed, with obstructive/SDB indications increasing over the study period. Obstruction/SDB now constitutes the primary indication for tonsil surgery in children, consistent with other published studies [27 28]. The incidence for infectious indications remained relatively stable over the study period.
The most common indication in the younger age group by far was obstruction/SDB, which was seen in this study as well as in several previous studies [10 28], likely reflecting an increased awareness of upper airway obstruction and SDB as a common disease in the pediatric population. In a single county study from Minnesota, a comparable large-scale shift in indications was observed with an increase in the percentage of SDB/obstructive indications from 12% in 1970 to 77% in 2005 [26]. Bhattacharyya et al. reported that the rates of paediatric adenotonsilletomy nearly doubled between 1996 and 2006, likely due to increased recognition of SDB and obstructive sleep apnoea (OSA) [29]. An increasing use of polysomnography (PSG) could have supported this theory of increased awareness of obstructive/SDB indications but no such data were available in our database for this cohort. Overall, PSG is very sparsely used in children in Sweden.
Another possible explanation to the increase in obstructive/SDB indications could be increasing prevalence of obesity. However, studies of obesity among Swedish children have observed an increase from 1984 to 2000, but thereafter a stabilization of the prevalence[30-32]. In our database no information about obesity was available and therefore this could not be further analysed.

Surgical methods
Along with a shift in indications, there has also been a shift in surgical methods, with TT gradually replacing TE as the most common surgical method for children in Sweden. This was also the conclusion of a study based on data from another Swedish registry independent of the NPR, the National Tonsil Register in Sweden [33]. Since 2011, TT has become more common than TE in Sweden. That same year, new national guidelines for TT were issued in Sweden [34]. One possible explanation for our findings could be that

Swedish ENT surgeons consider TT to be safe enough to perform on small children. TT has advantages over TE with respect to postoperative complications such as pain and bleeding [6 35] with comparable outcomes for symptom relief [36]. A known disadvantage of TT is regrowth of tonsils with recurrence of obstructive/SDB symptoms and need for reoperation. A recent Swedish study based on the NPR found a seven times higher risk for reoperation after TT than after TE, with the highest risk in the youngest age groups[37]. However, the clinical practice of TT for treating airway obstructions in children seem to be spreading; for instance, in Austria, several serious complications with posttonsillectomy haemorrhage has led to a switch to TT [5].

Strengths and limitations

In this study, there was a marked decrease in the incidence rate of tonsil surgeries between 1996 and 2001. The reasons for this decrease in the number of recorded surgeries were analysed together with the representatives from the NPR [38]. One reason for this dip could be a failure on the part of the NPR to collect data on performed surgeries, perhaps due to the increased use of day-care surgery between 1997 and 2001. Another reason could be insufficient reporting to the NPR from private hospitals, which became more numerous during this period. However, we believe this decrease was too great to be explained by failed registrations alone and must represent an actual decrease in the number of surgeries, although the true extent of this decrease cannot be precisely determined.

The NPR is generally considered to have high coverage [14], although like all clinical registers, it has potential inherent limitations, including inaccurate data reporting and missing values.

Major strengths include the large population size of the register and its wealth of unique nationwide data over a long time period. It can therefore be used to determine actual longitudinal application of a clinical practice in a specific country over long periods of time.

Conclusions

Our results showed considerable changes in clinical practices for tonsil surgery in Sweden during the period from 1987 to 2013. Overall, a doubling in the incidence rates was observed during this period. The reason for this change was an increase in surgical procedures due to upper airway obstruction/SDB, particularly among the youngest age group (1-3 years old) and among boys. Further, TT has gradually replaced TE as the predominant surgical method for obstructive/SDB indications. The incidence rates for infectious indications for tonsil surgery have remained relatively stable during the study period, and the most common patients in the infectious group were teenage girls.

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Footnotes

Ethics: This study was approved by the regional ethical review board of Gothenburg, Sweden (Dnr 534-14).

Contributorship statement: AB and JS contributed to the study design. AB, PN, DF, OS and JS contributed to the interpretation and analysis of data. AB wrote the first draft of the manuscript. PN, DF, OS and JS contributed to the final writing and revising of the manuscript, and checked for important intellectual content. All authors approved of the final manuscript as submitted.

Competing interests: None declared.

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Data sharing statement: No additional data available.

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38. Personal communication with Quality Manager for the Patient Register, Anders Jacobsson.

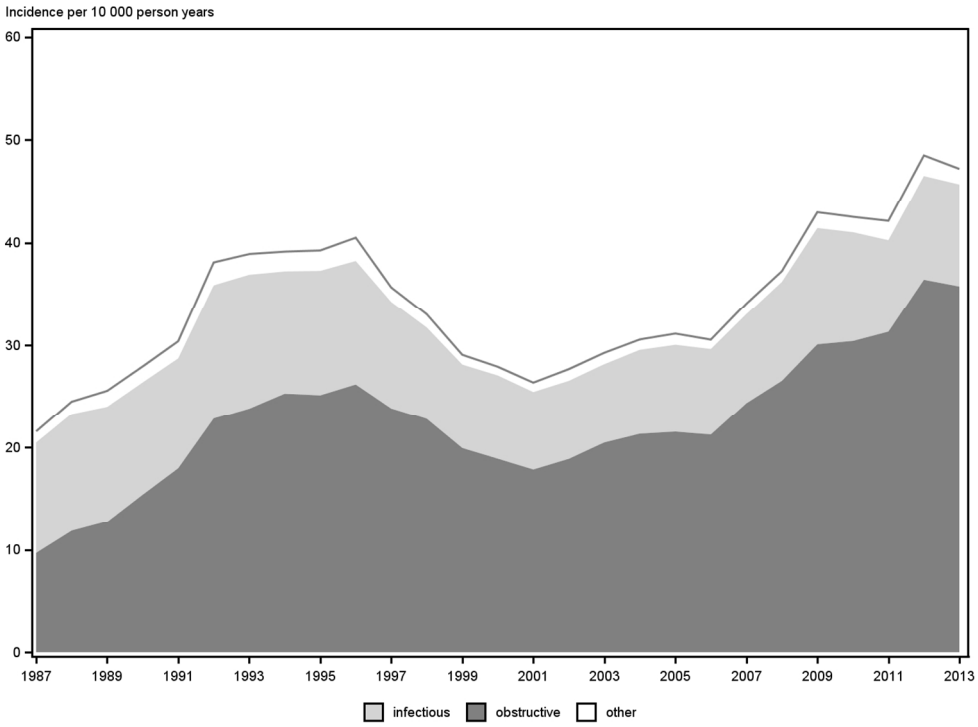

Figure 1. The incidence and indications of tonsil surgery in children aged 1-<18 years from 1987-2013. Incidence/10,0000 person years.

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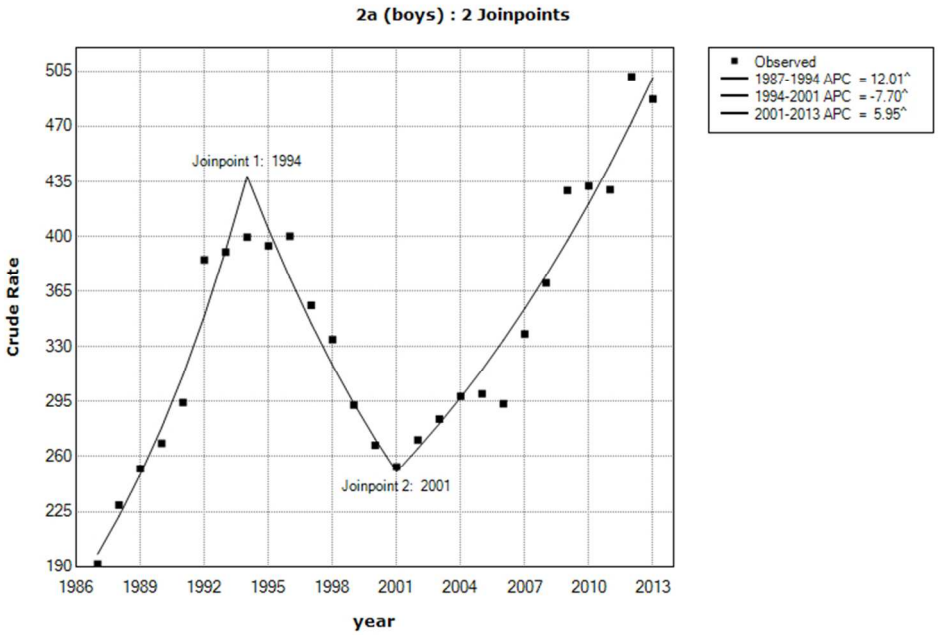

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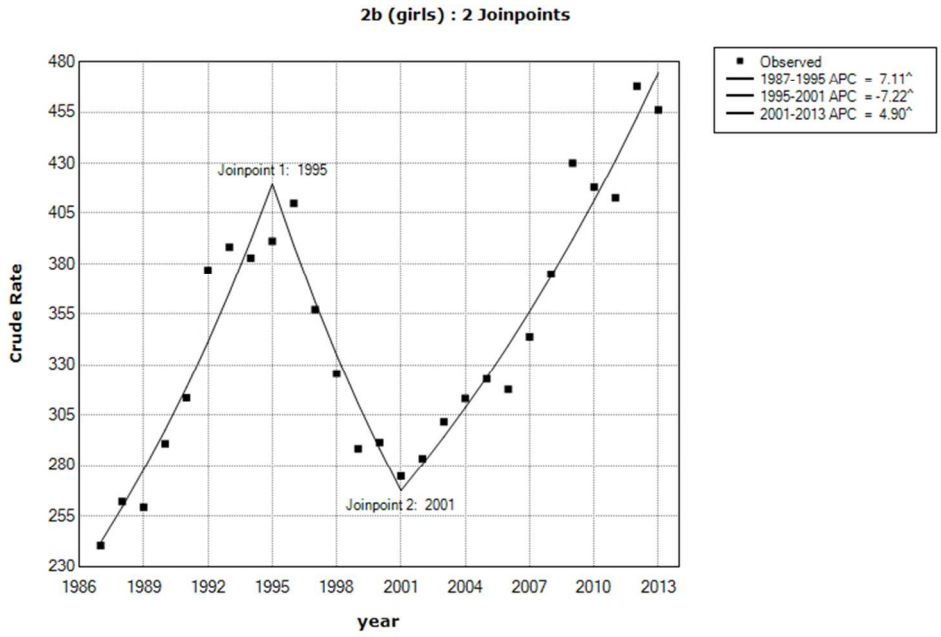

Figure 2 a+b. Joinpoint regression analyses showing the trends in the incidence rate of tonsil surgery for boys (2a) and girls (2b) 1987-2013. For both genders, two joinpoints were observed (1994 and 2001 for boys, 1995 and 2001 for girls). The crude rate is the incidence rate/100,000 person years. APC, annual percentage change.

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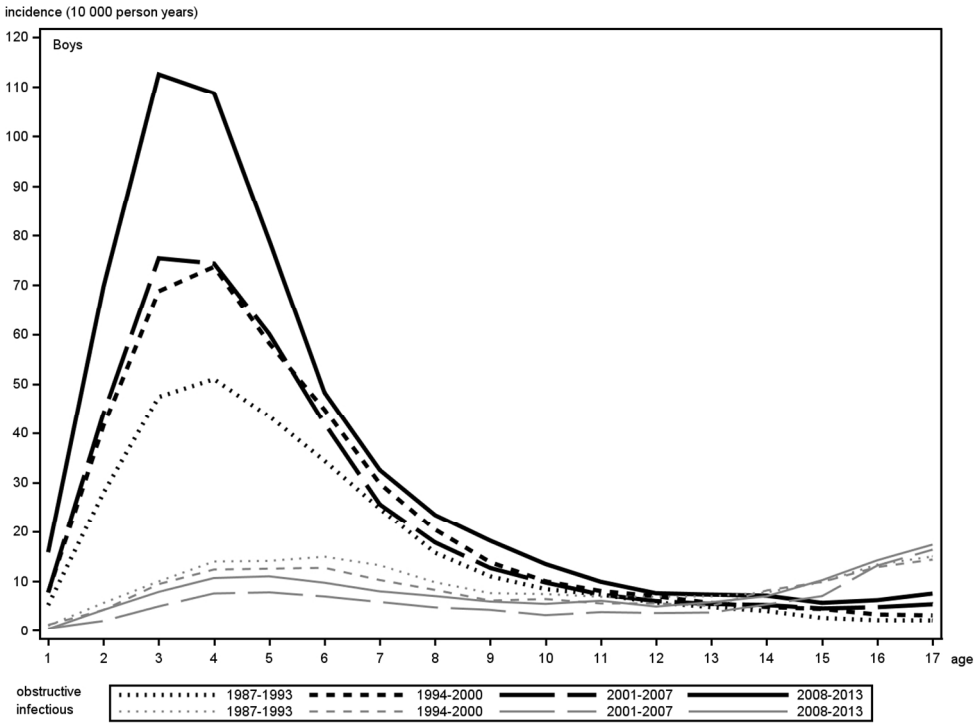

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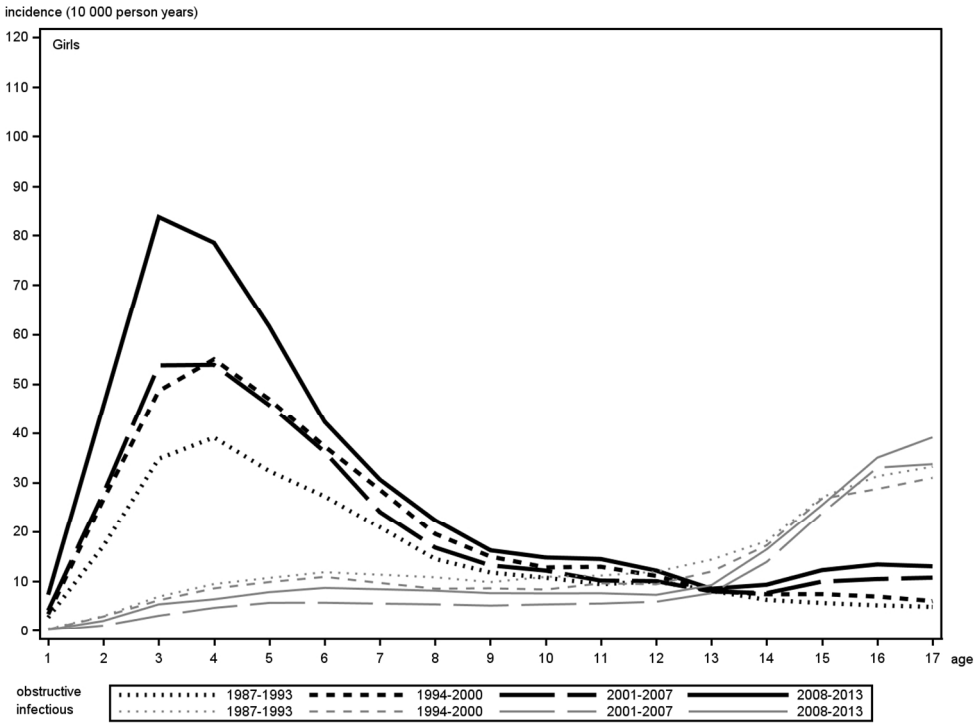

Figure 3a and b. Incidence of tonsil surgery procedures for boys (3a) and girls (3b) aged 1-<18 years between 1987 and 2013 separated by age and indication. Black lines represent obstructive indications and grey lines represent infectious. Each curve represents a 6- or 7-year period.† †

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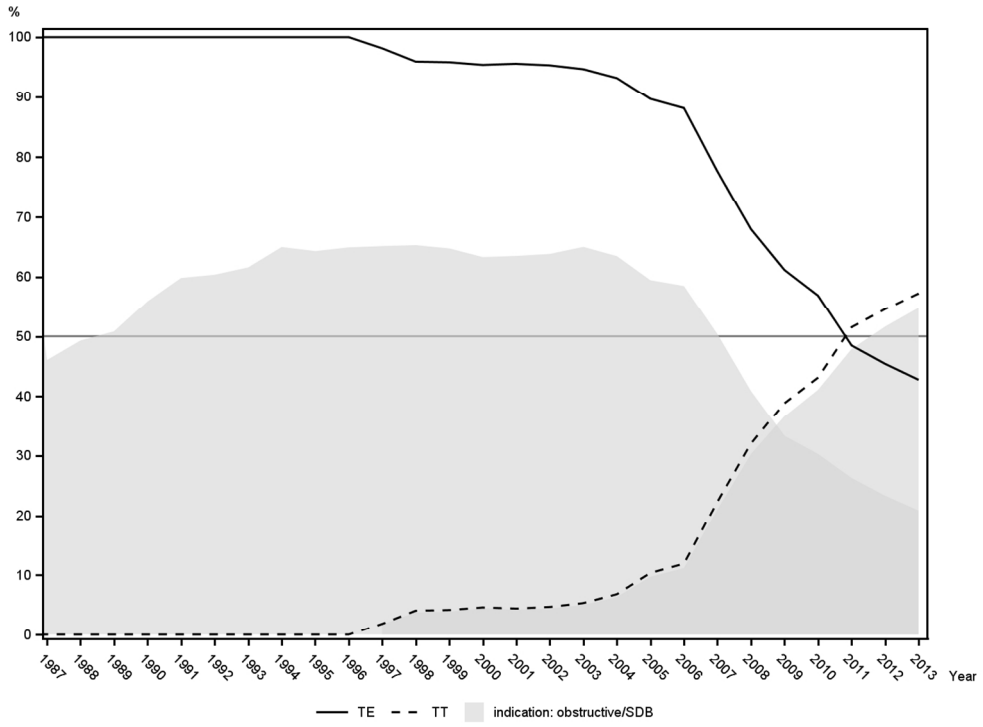

Figure 4. Distributions of the percentage of TE and TT procedures, with or without simultaneous adenoidectomy, performed between 1987 and 2013. The proportion due to an obstructive/SDB indication is shadowed for each method.‼ ┼
TE, tonsillectomy. TT, tonsillectomy.

119x90mm (300 x 300 DPI)

STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of *cohort studies*

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	1, 2
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	1, 2
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	2, 3
Objectives	3	State specific objectives, including any prespecified hypotheses	3
Methods			
Study design	4	Present key elements of study design early in the paper	3
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	3, 4
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up	4
		(*b*) For matched studies, give matching criteria and number of exposed and unexposed	-
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	4
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	3, 4
Bias	9	Describe any efforts to address potential sources of bias	
Study size	10	Explain how the study size was arrived at	4
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	5
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	5
		(*b*) Describe any methods used to examine subgroups and interactions	5
		(*c*) Explain how missing data were addressed	10
		(*d*) If applicable, explain how loss to follow-up was addressed	Not applicable
		(*e*) Describe any sensitivity analyses	Not applicable
Results			

Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	6
		(b) Give reasons for non-participation at each stage	Not applicable (register study)
		(c) Consider use of a flow diagram	-
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	6
		(b) Indicate number of participants with missing data for each variable of interest	Not applicable
		(c) Summarise follow-up time (eg, average and total amount)	Not applicable
Outcome data	15*	Report numbers of outcome events or summary measures over time	6, 7, 8
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	6, 7
		(*b*) Report category boundaries when continuous variables were categorized	-
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	-
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	7
Discussion			
Key results	18	Summarise key results with reference to study objectives	8
Limitations			
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	8, 9, 10
Generalisability	21	Discuss the generalisability (external validity) of the study results	10
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	11

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.