Revisiting systematic geographical variations in tonsils surgery in children in the Spanish National Health System: spatiotemporal ecological study on hospital administrative data

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ABSTRACT

Objective To provide new evidence on how tonsils surgery in children has geographically varied over time in the context of the Spanish National Health System.

Design Observational ecological spatiotemporal study on geographical variations in medical practice, using linked administrative datasets, including virtually all surgeries performed from 2003 to 2015.

Setting The Spanish National Health System, a quasi-federal structure with 17 autonomous communities (ACs), and 203 healthcare areas (HCAs).

Participants Patients aged 19 and younger residing in the HCAs and ACs.

Interventions Tonsillectomy with adenoidectomy (T&A); and tonsillectomies alone (T).

Main endpoints (1) Evolution of T&A and T rates; (2) spatiotemporal variation in the risk of receiving T&A or T surgery at regional level (ACs) and HCAs; and (3) the fraction of the variation (FV) attributed to each of the components of variation—ACs, HCAs, year and interaction ACs level.

Results T&A age-sex standardised rates increased over the period of analysis from 15.2 to 20.9 (5.7 points per 10,000 inhabitants). T alone remained relatively lower than T&A rates, evolving from 3.6 in 2003 to 3.9 in 2015 (0.3 points per 10,000 inhabitants). Most of the risk variation was captured at the HCAs level in both procedures (FV: 55.3% in T&A and 72.5% in T). The ACs level explained 15.5% in T&A and 17.5% in T). The ACs level explained 1.46% and 1.83% of the variation, respectively. The ACs year explained 1.46% and 1.83% of the variation, respectively.

Conclusion Our study showed widespread variations with a steady increase in rates and risk of T&A and a stagnation of T alone, where most of the variation risk was explained at HCA level.

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ This study covers virtually all the surgery provided to the population aged 0–19 in public settings in the whole country along years of observation; the cases have been effectively allocated to the place of residence so that the risks are unambiguously attributed to healthcare areas (HCAs), autonomous communities (ACs) and year.

⇒ A robust model spatiotemporal was used to describe the variation on different levels (ACs—locus of health policy decisions, HCAs (locus of provision) and over a long period of time (13 years).

⇒ A limitation of the study was the non-inclusion of surgical intervention in private hospitals with no public financing, although private provision in Spain is limited.

⇒ Baseline data analysis, influenced by factors both, at HCAs and ACs levels, but also on how unevenly ACs have adopted an evidence-based approach to tackle with unexpectedly high rates of tonsils surgery.

INTRODUCTION

‘In some of the early years of the School Medical Service it had seemed necessary to recommend further provision for the operative treatment of tonsils, but the tide rose so fast that in his Annual Report for 1923, Sir George Newman issued the first of his many warnings against premature resort to operation.’ This statement is part of the contextual arguments given to justify a dedicated session of the Section of Epidemiology and State Medicine at the Royal Society of Medicine on 28 May 1938. Sir Alison Glover, acting as speaker, provided evidence on differences in the rates of tonsillectomies that could not be warranted by differences in epidemiology. These proceedings, fairly deemed the first formal approach to the study of unwarranted variations in medical practice, paved the way for other works on the use of tonsillectomies in the paediatric population.
Numerous studies have shown wide geographical variations in tonsillectomy rates.4–10 As in Glover’s work, these large variations are suggested to be a proof of differences in care intensity and therapeutic choices across territories rather than differences in need. In Spain, a 2006 study based on 2002 data showed extreme variation in tonsillectomy rates, as large as 13-fold difference across 147 healthcare areas (HCAs), very unlikely to be explained by differences in children’ needs across HCAs.11

Since the 2006 study, no recent information on unwarranted variations in tonsillectomy rates in the Spanish National Health System has been released or published while guidelines,12–18 systematic reviews,19–23 randomised clinical trials24–28 and a cohort study29 on the indications for tonsillectomy and adenoidectomy multiplied. In the Spanish context, the societies of ENL and Paediatrics provided clear recommendations on the indications of tonsillectomy.30 There is a need to understand if this wealth of new evidence has translated into a change in the prescription patterns of tonsils surgery in children and consequently, into a change in the variation of population rates.

This paper provides new evidence on how tonsils surgery has geographically varied over time in the context of the Spanish National Health System.

Population
The population aged 19 and younger residing in all the 203 HCAs (the locus where hospital and primary care services are provided) of the 17 ACs (the locus where financing and policy-making decisions are taken) composing the Spanish National Health System was included.

Virtually all tonsillectomies with or without adenoidectomy performed on this population between 2003 and 2015 in all public hospitals in the country were included in the study. Tonsillectomy and/or adenoidectomy were defined as per ICD9-MC taxonomy. Codes 28.2 (tonsillectomies without adenoidectomy) and 28.3 (tonsillectomies with adenoidectomy) were considered for the analysis. All the tonsil surgeries identified were effectively allocated to the HCA and AC of residence.

Setting
The Spanish National Health System, composed of 17 AC health systems, covers virtually all the population residing in Spain for a wide and deep basket of benefits. Hospital care, outpatient specialised care, primary care and emergency are fully covered for free at the point of care, with no patients’ copayments. As a strongly regulated system, all the population is mandated to register to a primary care doctor (general medicine or paediatrics), and is allocated to an HCA where residents are served for specialised care. The private sector in Spain is essentially subsidiary; in 2015, the last year of our study, represented less than 5% of the total expenditure in health,31 with 85% of the hospital and outpatient specialised care provided in public settings.32

A usual pathway of care in the Spanish system would start from a visit to the primary care doctor who will decide on whether there is need for the patient to be transferred out to specialised care. In the case of tonsils surgery, the usual path for children with a tonsil condition would be as follows. The child will first visit the paediatrician at primary care level. Then, the paediatrician will decide whether the case requires transfer out to the ENL specialist. Usually, those ENL doctors specialised in children will provide the services to children transferred for all the HCAs in the region, and will decide on the treatment, whether watchful waiting, T&A or T alone. In the case of surgery, the intervention will be usually performed as a day-case care in dedicated hospital facilities or outpatient specialised care premises.

Main endpoints
Three endpoints were considered: (1) evolution of T&A and T standardised rates; (2) spatiotemporal variation in the risk of receiving T&A or T surgery at regional (ACs) and HCAs levels; and (3) the fraction of the variation attributed to each of the components of variation; so, ACs, HCAs, the year when the surgery was provided, and the interaction between the ACs and the year when the surgery was performed.
Sources of data

The study is based on administrative data from the so-named, Minimum Basic Hospital Discharge Dataset (CMBD in Spanish), that collects information from both in-hospital surgeries and day-case surgeries. Specifically, this data source provides administrative data (place of residence, provider where the population is allocated, and hospital of treatment); clinical information (main diagnosis, secondary diagnoses and procedures performed during the stay); and demographic information (age and sex) on all hospital discharges in the Spanish National Health System (public hospitals and private hospitals with public finding). The completeness of this data source is guaranteed since AGs have the legal obligation of duly reporting to the Ministry of Health, in a yearly basis, on the hospital activity (see the corresponding Royal Decree at https://www.boe.es/eli/es/rd/2015/02/06/69/com).

In addition, the denominator for the population rates was obtained from the annual census of the Spanish National Institute of Statistics. Atlas VPM allocates each of the episodes of treatment to the place of residence, then to the HCA and region of residence, both the units of analysis in this study.\(^\text{35}\) Notably, as part of its data quality assurance methodology, Atlas VPM conducts regular (once a year) quality checks to, for example, avoid over time coding variations, correct semantic and syntactic inconsistencies in the variables, or reallocate admission episodes to the place of residence if there are changes in geolocation of administrative areas or hospital providers.

Analysis

All 2003 to 2015 T&A and T interventions of patients aged 19 and younger were selected, linked to their area of residence and aggregated into four age-sex specific subgroups (0–4, 5–9, 10–14, 15–19). For each year in the series, age-standardised and sex-standardised rates per 100,000 inhabitants were calculated. As a first approach to variation the extremal quotient between standardised rates (95th to 5th percentiles ratio) and the interquartile interval (75th to 25th percentiles ratio) were calculated. Then, the systematic component of variation (SCV) provided the part of the variation that was not random (SCV assumes the expected cases for each HCA follow a Poisson distribution).

To understand the systematic differences in the risk of receiving T&A and T, risk ratios at HCAs and AGs level were estimated; thus, the variation across HCAs and AGs was decomposed in four components: spatial at HCA level, spatial at AC level, spatiotemporal at AC level, and overall temporal effect. Risks estimation and estimates for the fraction of variance followed a two-step approach: (1) under the hypothesis that risk remained constant across areas over time, the expected number of cases per HCA was estimated applying the overall rate over the period of study to the population at risk in each HCAs in the respective year. Standardised HCAs ratios (RR) were estimated using the ratio of observed-to-expected cases, interpretable as the maximum estimate of the risk ratio of admissions for T or T&A in each area in each period. (2) A Bayesian hierarchical approach was then used to model the different risk structures of dependence in both space and time.\(^\text{34–37}\) In the first level of this hierarchical modelling, we assumed that, conditional on the underlying relative risk, the number of counts \(y_{ijt}\) in the \(j\)th area at the \(t\)th time period followed a Poisson distribution with mean \(\theta_{ijt}=e_{ijt}\log(r_{ijt})\), where \(e_{ijt}\) is the number of expected counts and \(r_{ijt}\) the unknown relative risk. In the second level, the \(\log(r_{ijt})\) was expressed as the sum of the components representing the individual and independent contributions to the risk in a specific HCAs and period \(\log(r_{ijt}=\text{intercept}+S_i+T_j+ST_{ij})\), where the intercept term gives the initial level of risk that is shared by all regions and periods. The main effects \(S_i\) and \(T_j\) represented the additional risk of living in AGs \(i\) and period \(j\) and the second order interaction term \(ST_{ij}\) represented the risk contribution due to a combination of the effects that cannot be explained additively by the main effects. In the third level, a hyperparameter-prior distribution was assumed where the spatial effect was modelled following a convolution CAR prior.\(^\text{34}\) The temporal main effect was a combination of a time-unstructured (exchangeable) and a time-structured effects (first-order random walk), and the interaction term, as the independent effect of unobserved covariates for each combination of AGs and period \((ij)\), thus without any structure.\(^\text{35}\)

The use of a Bayesian approach in this study lays on a number of arguments. First, unlike in traditional studies on geographical variations, this study cannot assume that the exposure to tonsils surgery of the population residing in one area is independent of the exposure in neighbourd HCs; on the contrary, children from different HCAs get tonsils surgery at a single tertiary care setting that serves them all. On the other hand, traditional methods provide ecological associations (and interpretations) based on observed variables while latent unobserved factors that occur within the units of analysis are simply omitted; this Bayesian model allows inference on those latent unobserved variables that occur in units of analysis relevant to the study; so, we are able to know what part of the variation is in the regions (so, common to all areas), what part of the variation is in the HCA, and within the HCA, what part is structured (dependent on the interaction with other areas) or what is not (basically attributed to phenomena occurring exclusively within the area); and, what part is in the time trends (in the utilisation of certain procedures) and whether trends differ across regions. Finally, this Bayesian approach has been observed to be more robust in multiple testing conditions and more precise in the case of high heterogeneity across HCAs.

Patient and public involvement

Patients or the public were not involved in the setting of the research question, nor in developing plans for design/writing of our draft. Patients or the public were...
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*Figures are expressed in millions.
EB, empirical bayes; EQ5-95, 95th to 5th percentiles ratio; IQ25-75, 75th to 25th percentiles ratio; SCV, systematic component of variation; T, tonsillectomy alone; T&A, tonsillectomy and adenoidectomy.
not asked to advise on the interpretation or writing up of findings.

RESULTS

While the population 19 and younger increased by 8% until 2009, from 8.4M to 9.1M lives, remaining constant until 2015, tonsillectomies with or without adenoidectomies procedures grew at a faster pace: from 12218 in 2003 to 18110 in 2015 (48% increase).

T&A age-sex standardised rates steadily increased over the period from 15.24 to 20.96 (5.42 points per 10000 inhabitants). T rates in turn, lower than T&A rates, slightly increased over the period—from 3.57 to 2003 to 3.88 in 2015 (0.31 points per 10000 inhabitants) (table 1). By age groups (tables in online supplemental appendix 1), T&A were mainly performed in children 5 to 9 years old (46.8%) and 0 to 4 years old (35.6%), with an increase in T&A standardised rates of 8.11 and 7.63 points per 10000 inhabitants, respectively. T were more prevalent in the group between 15 and 19 (39.2%) and in children between 5 and 9 (33.5%), with an increase in T standardised rates of 1.42 and 0.59 points per 10000 inhabitants, respectively.

Figure 1  Surgery risk ratios across regions along the study period.

Variation statistics showed high variation figures; the systematic component of the variation was above 1.2 in all the period, slightly higher in the case of tonsillectomies alone (table 1). By age groups, the systematic variation in T&A was extreme; in the adolescents (15–19 years old), the SCV was between 0.68 and 1.2; in the group between 5 and 9, the SCV ranged between 0.5 and 0.63. In the case
of tonsillectomies alone, the systematic variation in these age subgroups was even larger; in the adolescents (15–19 years old), the SCV was between 0.75 and 1, while in the group between 5 and 9, the SCV ranged between 0.85 and 1.36 (tables in online supplemental appendix 1).

Figure 1 exhibits very large differences of tonsils surgery across ACs and over the study period, with larger variation in T&A.

Figures 2–4 show the spatial and temporal components of the variation in the observed to expected cases at AC level, and as HCA and AC jointly; in addition, it is also shown the average time evolution of the two prior models, for T&A and T alone (figure 2). As observed, T&A showed higher variation (9 out 17 ACs showed risk different to expected) while T showed a more homogeneous pattern of risks (only 3 out of 17 ACs had risk ratios slightly over or below the expected).

Figure 3 represents the joint model for both ACs and HCAs; the images in both T&A and T alone suggest that the higher contribution in the variation would correspond to the HCA. Finally, in figure 4, the time component shows a different pattern in T&A and T alone. While...
T&A keeps growing since the beginning of the study period, T alone has remained almost flat since 2006 (risk slightly above 1 with a slight increase in the last period).

Table 2 exhibits the relative contribution of each of these components of variation (ie, variance decomposition). Consistent with the observations in figures 2–4, most of the variance in T&A was captured by the spatial phenomenon, 55.4% explained by HCAs and 27.6% by ACs; the interaction between spatial and temporal components explained 15.6% of the variance while time just explained 1.5%.

In turn, the HCAs component explained 72.6% of variance in T (figures 2–4) with an additional 8.1% of spatial component of variance attributed to ACs level. The spatiotemporal interaction explained 17.5% of the variance while the temporal component explained a small 1.8% of the variation.

**DISCUSSION**

Our study showed a continuous increase in the rates of T&A procedures and a stagnation in T. The variability
was very high over the study period in both procedures, extreme in some age groups. The spatial component of the risk of surgery was the most relevant in the explanation of the variance, with HCAs capturing most of the variation (55% in T&A and 73% in T alone). In T&A, the variation attributable to the AC of residence was substantial, explaining a 27.6% of the variance, and notably different to T. The component of variance attributable to the spatiotemporal interaction was substantially relevant in T alone, explaining a 17.5% of the variation.

Variations in tonsils surgery are ubiquitous and wide. In our study, the extremal quotient was as high as 6.5 times in the case of tonsillectomies alone, and as 8 times in the case of T&A. Other studies exhibited a sevenfold variation in tonsillectomy rates across local authority areas in England in people under 15, 4.3-fold difference in USA and 3.4-fold in local areas in Veneto.

Likewise, our large variation in risk ratios and increasing intervention rates is also consistent with previous international and national evidence. Interestingly, the
behaviour is different between T&A and T. While the growing utilisation of tonsillectomies alone reached a plateau that has remained over the years, in the case of the combination with adenoidectomy has steadily grown. This relatively lower growth in T may be explained by the fact that there has been increasing evidence on the actual indications of this surgery as compared with the ambi-
fact that there has been increasing evidence on the actual
This relatively lower growth in T may be explained by the
behaviour is different between T&A and T. While the
combination with adenoidectomy has steadily grown.

As in previous research on geographical variations in
elective surgery in Spain, the variance component at the
HCA level has been observed more important than the
one at ACs level when explaining variation.37 This is consis-
tent with the institutional design of the Spanish Health
System and how decisions on provision are made; in this
particular case, ENT services are based and provided at
HCA level. However, this study has also found that the ACs
of residence explained a substantial part of the variation
when it comes to T&A (27.6% of the variation). As per the
analytical approach this finding would suggest that some latent unobserved factors common to all the HCAs
within an ACs would drive the decision of prescribing
T&A instead of conserving the tonsils and adenoids. A
potential latent variable (consistent with the observation
of greater rates of A&T in children aged 9 or less) that
would explain the high fraction of variation at ACs level is
that ENT specialised in younger children care, unlike the
rest of ENT doctors, provide de facto services at ACs level.

However, this explanation does not appear sufficient to explain that, for the overall period, some ACs are well
below the expected while others are well above (figure 2).
We may find a second potential explanation for these
differences across ACs in light of the importance of the
spatiotemporal component observed in our results (ie, 15.6% of the variance is explained by the interaction
between the ACs and the evolution of rates over time in
T&A). This interaction is coherent with the changes in the
indication profile over the years, uneven across ACs. So,
while in the first years of our time-series infections were
the underlying reason for surgery, currently, obstructive
conditions are those which drive the decision.

**Limitations**

Although the strengths of this work (i.e., the study covers
all the Spanish population aged 19 or younger, virtually
all the surgery provided in public settings in the whole
country along 13 years of observation and the cases have
been effectively allocated to the place of residence so that
the risks are unambiguously attributed to HCAs, ACs and
year) the study is not exempt from some potential bias.

As our study builds on administrative data, the spec-
ificity of codes and the coding practices are poten-
tial sources of bias, if recording differs across the units
of analysis. In the case of the Spanish National Health
System coding practices are fairly homogeneous, as the
Ministry of Health provides guidance since the late 1990s
on when and how to code the conditions and procedures
included in the International Classification of Diseases
(ICD). However, in the case of tonsils surgery ICD-MC
9th, the classification used until 2015, very specific in the
coding of tonsillectomy and/or adenoidectomy, does not
include any specific code for tonsillotomy, leading coders
to register this procedures as a tonsillectomy. Fortunately,
this fact, which would have implied a major limitation in
appropriateness studies, has not resulted in any bias when
it comes to the estimates of variation, since all tonsillo-
tomies were also included in the study.

As a second caveat, our analyses refer to the activity
carried out in public hospitals and private hospitals with
public financing. No information was included on those
private hospitals with no public financing. As private
hospitals in Spain do not distribute homogeneously
across regions, there is a potential for bias in the esti-
mates of variation and in the evolution of rates. There-
fore, our estimates are expected to be conservative in
those regions with larger provision of private services. If
we were accounting for those cases performed in private
hospitals in those regions, and those cases represented a
substantial amount, variation will likely increase at ACs
level although we cannot confirm the same for variation
at HCAs level.

As a third consideration, the strength of the results relies
on the large variation that is irrespective of age and sex
and the robustness of the estimates over time. But, when it
comes to the interpretation of the results, high (low) rates
or high (low) risk ratios cannot be strictly interpreted in
terms of adequacy—that is, more is necessarily worse, or
less is necessarily better. For that purpose, we would have
needed that the registry of the underlying conditions and
the encoding system (ICD 9th) would have unequivocally
flagged those cases deemed appropriate for treatment. As
this was not the case, we cannot make any strong judg-
ment about the right rate on tonsils surgery. However,
as this study builds on the whole population in mid to
large-size areas, we should not expect that the potential
differences in the prevalence of sleep-apnoea disorders
in children or severe infectious tonsillitis were as large as

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</table>

ACs, autonomous communities; HCAs, healthcare areas; T&A, tonsillectomy with adenoidectomy.
seven times, a variation observed in our series. This fact provides a fair indication that tonsils surgery rates are not necessarily driven by need.

As a fourth question, it would be ideal to continue the study until the most recent available data—2019. However, we prevented ourselves from continuing because of the discontinuation of the coding system in 2016 (ICD 10th started to be implemented) which entailed uneven coverage of cases across HCAs and AGs and severe data quality issues (eg, coding of secondary diagnoses). This is a limitation for the generalisation of our results nowadays, particularly if they were to inform any policy aiming the reduction of rates or the variation. Nonetheless, in an exploratory work carried out with a sample of HCAs from 2016 to 2018, the rates were observed to decrease but the variation in terms of extremal quotient remained very high, up to 7.

As a final comment, these findings have to be interpreted in the context of the Spanish National Health System—institutional design and care pathways. While the methodology is directly applicable to other contexts, the results (ie, the magnitude of the observed difference across providers, and the fraction of variation attributable to each level of analysis) may likely differ from one system to another. Thus, for a proper generalisation of the results to a different context, a judgement on the institutional design and usual care pathways will be required.

Implications
Spanish rates are low compared with OECD countries; among 30 OECD countries in the period 1994–2014, Spain was the 4th country with the lowest rates of tonsillectomies.9 However, variation still remains very high and a question on whether surgery is provided to eligible patients comes up when in our sample half of the patients receiving tonsillectomy alone and three in four patients getting both T&A did not have any condition deemed as an indication of surgery.

With the corresponding caveats out of these figures, while in the case of tonsillectomy alone, clear guidelines, proper antibiotic policies and audit and feedback mechanisms could reduce unnecessary variation in surgery, the lack of evidence about the use of T&A would indicate that policies should seek the reduction of the rates, for example, using, second opinion or disinvestment programmes. As a note for the potential impact of these policies, if those HCAs had reduced their T&A intervention at the level of the HCAs in the first decile of rates (0 rate could be sensibly assumed as inappropriate), the Spanish National Health System would have saved, over the study period, 121,535 interventions with a dubious indication.

As a final note of caveat, some evidence suggests that reductions in T&A rates may translate into an increase in hospital admissions for retropharyngeal and parapharyngeal abscesses or for acute tonsillitis.42 Before making any recommendation on substantial reductions in T&A rates, ENT doctors should propose guidance on those indications that would avoid those adverse events to appear.

CONCLUSIONS
Despite the rather low Spanish tonsils surgery rates, our study showed wide persistent variations with a stagnation of tonsillectomies alone and a steady increase in T&A rates, across HCAs and AGs, and over the years. Variation in the exposure to tonsils surgery could be explained by factors both, at HCAs and AGs levels, but also on how unevenly AGs have adopted an evidence-based approach to tackle with unexpectedly high rates of tonsils surgery.

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Contributors All authors contributed to conceptualising and designing the study. NM-L prepare the final dataset. JL and MC-M run the statistical analysis. MR, JL, MC-M, EA-P and SP commented on important intellectual content and made revisions. EC-R, MR and EB-D drafted the manuscript. All authors read and approved the final version of the manuscript. EC-R, MR and EB-D accept full responsibility for the work and/or the conduct of the study, had access to the data, and controlled the decision to publish.

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