"Thinking that somebody's going to delay [a tonsillectomy] for one to two years is quite horrifying really": A qualitative feasibility study for the NAtional Trial of Tonsillectomy IN Adults (NATTINA Part 2)

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Running title:

Feasibility study for the NAtional Trial of Tonsillectomy in Adults

Key words:

Feasibility, Tonsillectomy, Recurrent sore throat, Patients, Clinicians

Abstract

Objectives

Level One Evidence on the value of adult tonsillectomy versus non-surgical management remains scarce. Before embarking on a costly national randomised controlled trial, it is essential to establish its feasibility.

Design

Feasibility study with in-depth qualitative and cognitive interviews.

Setting

ENT staff and patients were recruited from nine hospital centres across England and Scotland.

Participants

Patients who were referred for tonsillectomy (n=15), a convenience sample of General Practitioners (n=11) and Ear, Nose and Throat staff (n=22).

Main outcome measures

1. To ascertain whether Ear, Nose and Throat staff would be willing to randomise patients to the treatment arms.
2. To assess General Practitioners' willingness to refer patients to the NAtional Trial of Tonsillectomy IN Adults (NATTINA) centres.
3. To assess patients' willingness to be randomised and the acceptability of the deferred surgery treatment arm.
4. To ascertain whether the study could progress to the pilot trial stage.

Results

Ear, Nose and Throat staff and General Practitioners were willing to randomise patients to the proposed NATTINA. Not all ENT staff were in equipoise concerning the treatment pathways. Patients were reluctant to be randomised into the deferred surgery group if they had already waited a substantial time before being referred.

Conclusions

Findings suggest that the National Trial of Tonsillectomy IN Adults may not be feasible. Proposed methods could not be realistically assessed without a pilot trial. Due to the importance of the question, as evidenced by NATTINA clinicians, and strong support from ENT staff, the pilot trial proceeded, with modifications.

Introduction

The role of tonsillectomy in managing adult sore throat remains uncertain, and despite demonstrable compliance with SIGN guidance [1], UK regional variation in tonsillectomy rates persist [2]. Questions that stakeholders wish to answer relate to the relative costs and benefits of tonsillectomy against non-surgical pathways. The 2014 Cochrane review [3] identified two evaluable adult trials, with just 156 participants, both in Finland, and concluded that reasonable levels of evidence on effectiveness were still only available for children. Low recruitment rates into surgical randomised controlled trials (RCTs) threaten external validity of findings [4]. Integration of qualitative research can improve feasibility, design and conduct [5]. Additionally, recruitment processes should be tested before patients are enrolled to a trial [4, 6] However there is a paucity of research examining key stakeholders' experience of recurrent sore throats and attitudes towards management in adults.

The NAtional Trial of Tonsillectomy In Adults (NATTINA) consists of this feasibility study, an internal pilot and definitive trial of 600 adults, with embedded qualitative process evaluation [7]. This paper, reporting the main findings from the feasibility study, follows a linked paper (Reference the linked NATTINA part 1 paper submitted separately) where stakeholders were asked their views of recurrent sore throat, tonsillitis and their management as part of this feasibility study. Gaining stakeholder perspectives of these issues was considered to be an essential part of the study, however the depth of findings allowed for two linked, but discrete papers to be completed.

In the main NATTINA trial participants will be randomly allocated into immediate or deferred surgery. Our experience of a randomised trial of tonsillectomy in children [8, 9], together with other published Ear, Nose and Throat (ENT) surgical trials [3], highlighted the problem of retaining participants in a non-surgical cohort. These findings along with patient and public engagement have influenced our trial design and decision to use deferred surgery as the conservative management option rather than no surgery.

The aim of the NATTINA feasibility study was to assess the practicality of the proposed internal pilot and full scale trial. The specific objectives of the study were to establish: standard NHS ENT staff willingness to randomise patients to the treatment arms; the feasibility of patient identification and the eligibility criteria ; GPs' willingness to refer patients to standard NHS NATTINA centres; patients' willingness to be randomised and their potential acceptance of the deferred surgery treatment arm, as well as views on the proposed data collection methods, including weekly sore throat alert prompts and Sore Throat Alert Returns (STARs)[7].

Methods

Ethical considerations

Favourable ethical opinion was given by proportionate review of the NRES committee – Fulham, London on 16 June 2014 (14/LO/1115).

Sampling

Sampling of patients was purposive, seeking maximum demographic (age/sex/duration/severity). A convenience sample was selected from NHS staff likely to be involved in the nine UK standard NHS NATTINA centres and GPs from the surrounding catchment areas. Sample size was determined by reaching data saturation where the research team deemed no new themes to have emerged in three consecutive interviews [10]. Based on previous work by the investigators [11], it was estimated that this plateau would occur at around 45+ interviews: 20 ENT staff, 15 patients and 10 GPs.

Procedure

ENT staff identified patients who met the proposed NATTINA eligibility criteria [7]. Healthcare professionals (otolaryngologists, research nurses, nurse practitioners, clinic managers and general practitioners) who were likely to be involved at each NATTINA main trial site were identified. Written informed consent was taken before interviews.

In-depth interviews took place over 5 months (August 2014 to January 2015) and lasted up to 30 minutes. Interviews were based on flexible topic guides derived from the literature, issues raised by our Patient and Public Involvement group and in conjunction with the study Otolaryngologists and GP. Themes and topics explored included: symptoms and effects of

recurrent sore throats, management of recurrent sore throat, experience of participation and willingness to participate in research.

Data management and analysis

Interviews were recorded and transcribed. Framework analysis [12] was supported by NVivo software [13]. Data were repeatedly read and coded by an experienced qualitative researcher LM within a framework of a priori issues and those identified by participants or which emerged from the data. Analysis was discussed at regular meetings of the research team to identify areas for closer consideration (including negative case analysis) and to enhance credibility of the thematic framework and interpretation [14]. Qualitative work explored influences on both patient recruitment and on the implementation of the study interventions. Analysis of barriers and facilitators to 1) trial participation and 2) the normalisation of study interventions in clinical practice was informed by Normalization Process Theory [15].

Results

All nine study centres participated, with 48 participants interviewed. Staff were 9 ENT consultants, 1 ENT trainee (registrar), 6 research nurses, 4 nurse practitioners and 2 trial managers. Seven centres received 39 patient 'expression of interest' forms yielding 15 (38%) patient interviews. Twelve patients were interviewed face-to-face and due to work commitments, 3 patients opted to be interviewed by telephone. At the time of their interview the patients were on the waiting list for a tonsillectomy; this was considered to be the most efficient form of recruitment. Contact details for 40 GPs were received from 7 of the centres; 11 (28%) GPs were recruited. All but one of the ENT staff and GPs were interviewed by telephone. Results are presented by study objective with individual participant quotations used to support and illustrate the findings.

<u>ENT staff willing to consider participation in NATTINA</u>

All interviewed staff were willing to participate in the NATTINA trial and to randomise patients, however, they questioned whether patients would be willing to accept randomisation:

> *We don't know which arm you're better off being in. So I'm very happy randomising the patients. I don't know how acceptable it is to the patients we randomise*

Most ENT staff felt the research would address the fundamental question of whether tonsillectomy in adults was beneficial, not only to the patient but also for the NHS in terms of cost-effectiveness.

I mean, whilst we've driven down the frequency of tonsillectomy and the so-called savings there, what we do see is an increase in people coming into hospital with acute tonsillitis

The above respondent felt the implications of these 'so-called savings' were having a negative impact on patients' health. Many ENT staff felt that the evidence for surgery versus conservative management was scarce:

..A lot of decisions are being made about how to treat patients with recurrent acute tonsillitis which don't have a robust evidence base behind them

It was felt that new research evidence had the potential to improve patient care and practice.

<u>Potential participants willing to take part in NATTINA</u>

Patients' views

The majority of patients reported that they would not consider taking part in the proposed trial as they did not want the risk of being randomised to deferred surgery:

..I'd be anxious to have the surgery sooner because I've been suffering since I was young...to wait even more and to miss more time off work, no I really think it's time that they come out

Many patients reported the negative effects of tonsillitis and felt that to defer surgery would be too detrimental to their quality of life:

It had too much of an impact. It was happening at least twice a month as well, so it was really interfering with my attendance and stuff, and work, and money

Some patients stated they might consider randomisation if they knew they could opt out of the deferred surgery group without having to go back to the bottom of the surgery waiting list if their symptoms worsened:

Yeah, that would probably be better. I wouldn't want to be in the position where you have to wait and wait and wait to get re-referred and re-seen and stuff like that

GPs' views

Many GPs believed that tonsillectomy in adults was a rare occurrence and they were under pressure to minimise referrals to secondary care. Some felt that adults were not looking for surgery:

We try really hard not to send our patients because for the vast majority of patients they are unlikely to have their tonsils removed as adults

However, those GPs that did refer patients felt the patients would probably be unwilling to be randomised as, having had to meet strict criteria before being referred[1], GPs felt that by the time the patient reaches the consultant they have an expectation of surgery:

Certainly, I do not refer very many people, and the ones that I do, I do not think they would be willing to say "Great, I will enter a trial and wait up to another two years"

As with the actual patients themselves, a couple of GPs reported that the option to change to immediate surgery if withdrawing from the deferred surgery group may persuade some patients to be randomised:

I think that sounds very reasonable. I like the get out clause in the control arm, but I think it's a very good idea to delay surgery anyway

ENT staff views

Most ENT staff concurred with GPs' views of patients having expectation of surgery, not willing to defer surgery, or that they may be more willing to be randomised with a quicker 'opt-out' route:

Most patients in my experience do come with the view point that they would like the tonsils removed 'cos a lot of them have already discussed it with the GP. So asking them to wait another year, I'm not sure if we'll be able to recruit that many patients to that arm

Treatment pathways acceptable and adequately defined

Patients' views

Participants' reactions were closely related to whether they would be willing to be randomised and take part in the trial. That is, they were not willing to accept the risk of being randomised into the deferred surgery group as they found that pathway, as originally presented, to be unacceptable:

> *I don't even want to imagine what that would be like [deferred surgery]. Tonsillitis is honestly one of the worst illnesses I've had, and needs to be treated sooner rather than later.*

GPs' views

A few GPs felt that deferring surgery was a good idea but were unsure how it would be accepted by patients:

> *I think that's entirely appropriate. My only concern is the non-surgical treatment plan you might lose patients who then go on to decide that they want surgery… because they are having difficulty with their symptoms*

ENT staff

Some ENT staff identified concerns over the treatment pathways with one practitioner worried that patients may feel they were being disadvantaged by being randomised to the deferred surgery arm; one research nurse felt that delaying a patient's surgery was not acceptable:

> *Thinking that somebody's going to delay that [a tonsillectomy] for one to two years is quite horrifying really*

Outcome measures and data collection methods feasible and adequately defined

Patients' views

Most patients found the outcome measures and data collection methods acceptable with most happy to use electronic methods (email and text) to communicate and complete tasks. A small selection of patients reported that they preferred paper-based methods but understood that email would be quicker and cheaper.

Patients reported that they would be willing to complete the STAR text alerts, however some felt weekly to be too onerous:

I'd probably get quite irritated after a while like weekly seems just too close together. Maybe like fortnightly would be a better idea

ENT Staff views

Some staff felt that data collection was an on-going research issue and that some patients may find the methods intensive. Paper-based methods were stated as rarely successful and there was concern weekly alerts may be too burdensome.

However, there was general consensus that electronic methods would be suitable:

The demographic of recurrent tonsillitis tends to be younger patients, so I think things like text messaging questionnaire [sic], etc. would probably give you a higher response rate than a traditional paper-based through the post questionnaire.

However, one staff member was concerned that not everyone would have access to the internet. Additionally it was suggested that patients, not currently suffering symptoms, would be disinclined to respond to prompts for outcome data. Furthermore, staff from two of the centres reported high numbers of patients whose first language was not English:

The only issue is the language barrier for some patients, which where I work, my patient population, that's quite a big issue.

<u>Process of patient identification and recruitment feasible and adequately defined</u>

GPs' views

Most GPs stated that they were willing to refer patients to centres participating in NATTINA but there were some queries about how the referral process would work:

It is maybe thinking about, in terms of the study design, people at the point of referral knowing, or the point they receive their outpatient at clinic is probably better…that gives them the chance to almost revisit why they have been referred, and what their expectations are .

Many GPs thought it beneficial for practices to be study aware so they could provide patients with information to ensure that patients are referred without a prior expectation of tonsillectomy.

Discussion

Synopsis of key findings

Results suggest that ENT staff are strongly supportive of a trial of tonsillectomy in adults and are willing to randomise patients meeting SIGN criteria[1] . However patients meeting NATTINA eligibility criteria expressed reluctance to be randomised because of unwillingness to enter the deferred surgery arm of the study. Patients indicated that they might be more willing to be randomised if there was a clear route back to surgical intervention - at the time of their interview most patients had received a surgery date for their tonsillectomy or one was imminent. This inevitably contributed to their negative perception of deferral. There is a lot of emotion associated with surgery and, whilst waiting, patients are often preoccupied with issues such as feeling 'in limbo', 'lives being on hold' and 'clock-watching'[16, 17].

Many GPs believed tonsillectomy in adults was a rare occurrence. However, in 2013-14, 20,607 adults over the age of 16 years received a tonsillectomy in England i.e. the average GP will refer 2 patients (who receive a tonsillectomy) every 3 years [18]. In this study it was reported that some GP practices were encouraged to minimise tonsillectomy referrals; it has been estimated that two thirds of Clinical Commissioning Groups restrict referrals for treatments they deem to be non-urgent or of low clinical value [19]. This means that treatment control pathways have changed, moreover, some GPs stated they very rarely saw patients who were eligible for referral.

Implications for pilot trial

The feasibility trial allowed for timely modifications and valuable stakeholder insights. The Trial Management Group assessed the feasibility results and implemented several changes, specifically around movement between treatment arms. The proposal that patients who wanted to switch from the deferred surgery group could do so without going back on the waiting list resulted in positive feedback from patients. This proposal arose from a research team meeting to discuss interim analysis of the feasibility study and feedback from the patient involvement panel. It was proposed that reduced waiting for

patients who decide to switch may be enough of an incentive for some patients to participate. Random allocation to treatment arms within NATTINA will be concealed from investigators, GPs, ENT staff and participants in order to eliminate bias however, anything which facilitates movement between arms has the potential to impact on the intention to treat analysis; therefore, it was recommended that a per-treatment analysis was also conducted and that numbers switching are monitored throughout the recruitment period to assess impact on trial design. The following changes were also recommended:

- Emphasis on the need for a trial in the patient information materials
- Spread the recruitment to the pilot to a larger number of centres
- Refinement of baseline questionnaire
- Translation of patient study information to Urdu, Gujarati, Punjabi and Bengali
- Clarification of clinical pathway for control (deferred-surgical) arm for participants
- Extra attention to dissemination of information about the study to GPs, to mitigate patient expectation that referral equates to tonsillectomy

Strengths and limitations

A unique strength of this study is the quantity of appropriately representative data from multiple stakeholders. However, the fact that we selected patients who had already decided to proceed with a tonsillectomy inevitably must have influenced their perception of the study.

Conclusions

The proposed methods were generally acceptable notwithstanding some concern about the weekly frequency of sore throat episode recording. ENT and research staff stated that the acceptance of the data collection methods could not realistically be assessed until a pilot trial was in operation. A decision-making meeting was scheduled for the end of the feasibility study to review the findings and to confirm that there was sufficient support from those interviewed to allow the project to continue on to the NATTINA internal pilot phase. The decision to continue was approved by the NATTINA Trial Steering Committee and HTA informed. Barriers to recruitment which may emerge include: fewer eligible patients than expected, smaller percentage of patients agreeing to participate, internal staff problems, [20] and lack of equipoise [21].

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