Treatment options for patients with pilonidal sinus disease: PITSTOP, a mixed-methods evaluation

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Disclaimer: This report contains transcripts of interviews conducted in the course of the research and contains language that may offend some readers.

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Scientific summary

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Scientific summary

Background

Despite being a common condition, there is no clear consensus as to how pilonidal disease should be managed. Throughout the UK, surgical practice is varied, and existing literature largely consists of single-centre cohort studies using a range of disease classification systems, interventions and outcome assessments. There is a need to improve clinical management of this condition and define future research processes and priorities.

Objectives

PITSTOP aimed to investigate surgical options for the treatment of pilonidal sinus disease (PSD). The objectives were to:

- Follow patients with symptomatic pilonidal sinus referred to each collaborating site, prospectively recording details of their pit/track anatomy, surgical management, medical events and health-related quality of life (QoL) until 6 months after their operation.
- Describe the combination of interventions currently in use and quantify clinical and patient-reported outcomes (PROs) associated with each.
- Identify patient-specific disease features that might predict poor outcome in each treatment group by risk-modelling methods.
- Derive a case mix-adjusted estimate of the risks associated with common treatment options, using causal inference methods to provisionally rank the optimal management strategies among patients for whom more than one treatment is considered appropriate.
- Provide an overview of patient views and experiences.
- Collect the views of patients on which interventions they would rather avoid and which outcomes they most value.
- Reach a surgeon-based consensus on which subtypes of pilonidal disease may benefit from which treatment options.
- Reach a surgeon and patient-based consensus on research priorities.

Design and setting

PITSTOP was an observational cohort with nested mixed-methods and qualitative design which took place across 33 NHS Trust sites in the UK. The study had an additional clinician survey component and validation of a classification system and culminated in a three-stage Delphi exercise to identify research and practice priorities.

Participants

Eligible patients undergoing surgical management for PSD and interested in participating were consented to the study. Participants referred to a collaborating centre for definitive elective surgical treatment of PSD were required to meet the following criteria:

- Consenting patients over 16 years of age with PSD.

Participants were ineligible if any of the following conditions were met:

- Asymptomatic
- Pregnant
- Unable to give consent
- Acute abscess
- Hypersensitivity to the sealants.

Intervention

This was a non-intervention study with the choice of treatment being a shared decision made between the surgeon and patient. Treatment of PSD typically comprises two essential components (with the exception of phenol injection, seton and fistuloscope/diathermy – which aim to induce fibrosis with or without obliteration of the tracks):

1. Excision of the affected skin and fat (the amount of which varies among patients, and surgeons differ with respect to how the resultant wound is managed).
2. Closure of the wound, which is either left open and heals slowly by secondary intention or closed using sutures, also known as primary wound closure.

There are 18 excision–closure combinations that are theoretically possible. Therefore, the operative intervention was recorded by the following excision and closure techniques:

Excision procedure

- Local excision.
- Curettage.
- Phenol injection.
- Pit picking.
- Seton.
- Laser-assisted excision.
- EPSiT (endoscopic pilonidal sinus treatment).

Closure procedure

- Primary midline closure.
- Fibrin glue (FG).
- Marsupialisation.
- Lateral closure (Karydakis; Bascom's cleft).
- Flap (Limberg; rhomboid).

Follow-up

Participants were followed up 1 day and 7 days after surgery (with day 0 the day of elective surgical treatment), at the routine clinic visit appointment, at 6 months, and at the end of study.

Main outcome measures

As PITSTOP is a cohort study examining current practice, there are no primary outcome measures. The following data were collected:

- pain (numeric rating scale) on day 1 and day 7 postoperatively and at each follow-up
- EuroQol five dimensions five levels questionnaire (EQ-5D-5L) QoL at each follow-up

- interactions with primary and secondary care
- length of time to healing
- return to normal activities
- complications
- recurrence
- infection.

Statistical methods

The study aimed to recruit 800 patients, with at least 100 within each of the front-running management strategies. Two treatment comparisons were undertaken: any major excisional procedure versus any minor procedure, and asymmetric closure versus any minor procedure (minimal excision).

Regression modelling, propensity score-based approaches [inverse probability weighting (IPW) and nearest neighbour matching] and augmented IPW were used to account for measured potential confounding. Continuous outcomes (pain at day 1 and day 7) were modelled using linear regression; binary outcomes (recurrence, complication) were modelled using logistic regression, with marginal probabilities used to estimate absolute risk difference. Time to wound healing and time to return to normal activity were modelled using parametric accelerated survival time.

The following features were considered as potential risk factors for outcome and treatment choice: sex, body mass index, depth of natal cleft, presence and type of gluteal hair, smoking status, pit density, presence of unilateral or bilateral disease, distance from furthest lateral opening to the nearest pit, presence of pus and Wysocki disease classification. Features affecting treatment choice were assessed using logistic regression with treatment choice as the outcome, and the same propensity score adjustments were used for each outcome. For each outcome, three regression models were fitted: one that adjusted for all listed features, one that adjusted for features associated with the outcome (based on Akaike's information criteria, model *c*-statistic and clinical review) and one that adjusted for disease classification alone. The difference in predicted outcome and corresponding 95% confidence interval (CI) were estimated for each modelling strategy on each outcome.

Mixed-methods case studies

Mixed-methods case studies were undertaken to understand why people make and regret decisions on their treatment. Longitudinal semistructured interviews (20 cohort participants from 13 sites) were conducted at baseline and 6 months later with framework analysis mapped findings to Witt's coping in deliberation framework and Sekhon's acceptability framework. We triangulated findings with baseline scores CollaboRATE shared decision-making (SDM) survey scores and 6-month decision regret (DR) scores.

Discrete choice experiment

An online survey using the discrete choice experiment (DCE) method was used to ask patients to choose their favoured treatment when presented with competing hypothetical treatment profiles. Regression analyses were conducted on DCE responses.

Clinician survey

A survey was developed following the CHERRIES statement checklist of recommendations to identify the most frequently used interventions for specific clinical scenarios in current PSD practice. It was

disseminated via the surgical trainee research collaboratives to practising consultants throughout the UK. Data were captured and stored in the REDCap software.

Consensus exercise and validation of a classification system

Two separate surveys were undertaken. The first comprised a Delphi consensus exercise in which surgeons and patients were asked to recommend best practice and further research. In the second, 15 surgeons were each asked to retrospectively assess photographs to quantify agreement in the Wysocki classification tool, with 90 patients each assessed by 6 surgeons.

Results

Cohort study

Participants

Thirty-one UK sites recruited participants over a 46-month period from May 2019 to March 2022. Seven hundred and twenty-nine participants consented to the study; after exclusions due to no procedure (*n* = 45), incorrect diagnosis (*n* = 7) and insufficient treatment information (*n* = 10), there were 667 participants included in the analysis cohort. Six-month follow-up data were available for 71% of participants; recurrence and complication data were available for 94% and 96% of participants, respectively.

Main results

Sixty per cent of patients (*n* = 397) received a major procedure; this comprised 272 (41%) asymmetric closure, 49 (7%) leave open and 76 (11%) midline closure. The remaining participants received minimal excision (*n* = 270, 40%), most commonly glue (*n* = 106, 16%) or pit picking (*n* = 60, 9%). Pain on day 1 and day 7 was higher for patients that received major procedures compared to minor procedures (augmented IPW-adjusted mean difference in day 1 pain 1.58 points, 95% CI 1.14 to 2.01 points, *n* = 536; mean difference in day 7 pain 1.53 points, 95% CI 1.12 to 1.95 points, *n* = 512). The difference was broadly consistent regardless of adjustment method.

Complications were reported by 207/385 (54%) participants that had major procedures, and 94/258 (36%) participants that had minor procedures (augmented IPW-adjusted risk difference 17.5%, 95% CI 9.1 to 25.9%, *n* = 579). Recurrence was reported for 86/373 (23%) of major and 87/256 (34%) of minor procedures; the augmented IPW-adjusted risk difference was −10.1% (95% CI −18.1 to −2.1%, *n* = 575). The estimated difference between groups was smaller for treatment failure (a composite of recurrence, failure to heal, and failure to return to normal activities), where 45% of major procedure participants versus 47% of minor procedure participants experienced treatment failure (augmented IPW-adjusted risk difference −2.3%, 95% CI −10.9 to 6.2%). The estimates for treatment difference in recurrence and complication were consistent across the various adjustment methods.

Treatment differences for time to healing and time to return to normal activities were less consistent between adjustment methods. Participants receiving major procedures took an estimated minimum of 20 days longer to return to normal activities (augmented IPW difference 25.9 days, 95% CI 18.4 to 33.4 days). Difference in time to healing was estimated to be at least 34 days more in the major group, but the estimates and corresponding CIs were inconsistent across models. At 6 months, around 25% of participants in both groups had wounds that had not healed; 12% of major procedure participants and 4% of minor procedure participants were yet to return to normal activity.

Comparisons of asymmetric closure with minimal excision produced results similar to the major versus minor comparisons.

Clinician survey

The survey was completed by 109 participants (54.5% response) who routinely cared for patients with PSD. Respondents reported a median caseload of 15 patients per year [interquartile range (IQR) 10–20]. Of those estimating their recurrence rates, 28.8% predicted they were in the 16–30% range. A wide range of treatment strategies were employed, with 65.1% practising an excision-and-leave-open technique despite this being considered obsolete as a treatment option. Surgical training in this obsolete technique was experienced by 83.1%. Many recommended non-surgical treatments despite the lack of evidence.

Mixed-methods case studies

No choice of treatment was offered to 9/20 participants, although this was not always seen negatively on the CollaboRATE instrument. Decision-making was influenced by prior experience of pilonidal sinus and anticipated recovery time. Participants scoring highest on DR also had among the highest SDM scores. The burden involved in wound care and the gap between expected and actual time for recovery were the principal reasons given for DR.

Discrete choice experiment

The survey was completed by 111 participants. The strongest predictor of treatment choice was risk of infection/persistence (attribute importance 70%), followed by shorter recovery time (attribute importance 30%). Patients were willing to trade off recovery time against risk of infection/persistence. Patients aged over 30 years had a higher risk tolerance (22.35–34.67%) for treatment failure if they could experience rapid recovery. Younger patients were willing to accept smaller risks (maximum acceptable risk 1.51–2.15) in exchange for a faster recovery. Patients were uniform in rejecting excision-and-leave-open because of the protracted nursing care it entailed.

Wysocki tool classification

The Wysocki tool demonstrated acceptable agreement. Overall, the kappa statistic for agreement was 0.52 (95% CI 0.42 to 0.61), with five of the six surgeons reaching consensus in 53 (59%) patients. Agreement did not differ with regard to the surgeon's experience. This, along with the prognostic value of the tool, suggests this could be used in practice to classify PSD.

Consensus exercise

The top five practice statements included: any treatment should not be worse than the disease itself; minimally invasive procedures should be preferred when feasible; surgeons should have opportunities to learn new techniques; a classification tool will help inform treatment options; and delayed return to activities is an important outcome.

The top five research statements included: future trials should compare broad groups of operations – minimally invasive procedures versus major excisional techniques, with stratification by severity of disease; a core outcome set and PROs should be used; there should be an attempt to develop an algorithm or decision tree to aid surgeon decision-making.

Conclusions

The burden of surgery for PSD is significantly greater than that reported in the literature. Many surgeons perceive this but continue to practise outdated procedures. While minimally invasive procedures may reduce this burden in many patients, they are not always offered when they could be. This practice may be driven by the desire to achieve cure at the expense of protracted recovery, but this is not always what patients want. Many would trade reduced chance of cure for more rapid recovery. Future practice priorities should follow the ethos of not making the surgery worse than the disease itself and appreciate that patients need to be properly informed about the burden associated with wound care and the likelihood of recurrence associated with different procedures, to minimise DR. Future trials should compare broad groups of interventions (minimally invasive vs. major excisions) stratified by disease severity and utilising a reliable and validated Wysocki classification system. Such trials should incorporate a core set of PROs.

Trial registration

This trial is registered as ISRCTN95551898.

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Study protocol

The original protocol for the PITSTOP study can be found here. https://figshare.shef.ac.uk/articles/journal_contribution/The_PITSTOP_Study_PIlonidal_sinus_Treatment_STudying_the_OPtions_-_Protocol/7578242. A list of the protocol amendments since the initial Research Ethics Committee (REC) approval can be found below.

Version number	Changes made	Date of REC/ HRA approval	Amendment number
1.2	Errors in the flow diagram Errors that were overlooked such as age where it stated 18 years instead of 16 years Typological errors in the text Changes to the call at day one post surgery – this is now a phone call by a research nurse	03 May 2019/03 April 2019	SA01
1.2	Changes of collaborator Christine Moffatt University affiliation – previously University of Nottingham, now Nottingham Trent University (p7)		
1.2	Addition of optional consent item described which is the taking of photos pre-surgery to assist with the development of the classification system for PSD (p19)		
1.2	A follow-up call on day 1 post surgery as opposed to a text message (p19, p34, table 1)		
1.2	Removal for the 4–6 week time frame from routine clinic visit follow-up as this varies from clinic to clinic (p34)		

Version number	Changes made	Date of REC/ HRA approval	Amendment number
1.2	Addition of infection as outcome measure (p33)		
1.3	Clarification: Patients must have a minimum of 24 hours between receiving the Patient Information Sheet and consenting to the study (p16)	N/A/19 November 2019	MA07
1.3	Clarification: The postoperative routine clinic visit may also be conducted by the telephone if considered routine at site (p19)		
1.3	Clarification: The recruitment end date is July 2020 (p29)		
1.4	An additional telephone follow-up can be made if the participant does not attend the pre-arranged face-to-face clinic visit (p19)	N/A/03 December 2019	MA08
1.4	Clarification: Data can be collected by trained research personnel or a delegated member of the research team (p19)		
1.4	Addition of the REC reference on the front page (p1)		
1.5	Clarification: The photographs will be used to aid the validation of the classification system for PSD (p19)	02 March 2020/07 April 2020	SA03
1.5	Clarification: Preoperative photographs of the surgical site will be uploaded on the REDCap data capture system (p20)		
1.5	Clarification: A copy of the consent form will be uploaded on to the REDCap data capture system for monitoring purposes (p20)		
2.0	Clarification: Sponsor and CI e-mail addresses have been updated (p6)	03 August 2020/05 August 2020	SA04
2.0	Clarification: Study coordinating staff list has been updated (p8)		
2.0	Response to COVID-19: Addition of postal consent if preoperative consultations are conducted remotely OR social distancing measures inhibit research personnel to gain consent in person (p15)		
2.0	Surgeon survey is also accessible via an online link hosted on REDCap (p18)		
2.0	Research personnel may text a participant to arrange a follow-up call (p18)		
2.0	Response to COVID-19: Baseline measures can be taken over the phone (p19)		
2.0	Response to COVID-19: Follow-up data can be collected by study coordinating team if research personnel at NHS trust do not have capacity (p19)		
2.0	Response to COVID-19: missed 6-month follow-up data can be collected at the end of study (p19)		
2.0	The DCE questionnaire can be completed by non-PITSTOP patients. The questionnaire can be advertised using a leaflet and/or social media (p24)		
2.1	Research personnel may e-mail a participant to arrange a follow-up call (p18)	N/A/27 October 2020	MA11
2.2	Update to study coordinating team (p7)	N/A/29 September 2022	NSA13
2.2	Clarification of study procedures for the PSD seton surgical procedure (p19)		

Version number	Changes made	Date of REC/ HRA approval	Amendment number
2.2	Clarification of follow-up procedures (p20)		
2.2	Update to recruitment end date (p30)		
3.0	Amendment to consensus technique (p25)	18 November 2021	SA05

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This article

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