

Clinical effectiveness and costs of the Sugarbaker procedure for the treatment of pseudomyxoma peritonei

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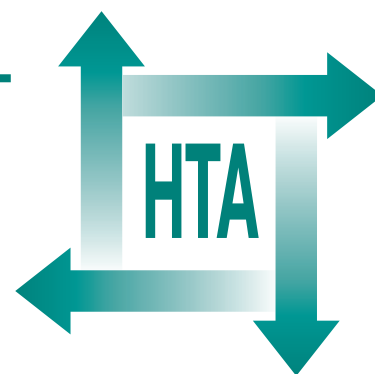
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Executive summary

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Executive summary

Epidemiology and background

Pseudomyxoma peritonei (PMP) refers to a progressive disease process within the peritoneum, thought to originate in the appendix and characterised by the production of copious amounts of mucinous fluid resulting in a 'jelly belly'. If untreated the condition is fatal. Uncertainty persists as to the specific definition, pathology, site of origin and prognosis of PMP. It is a rare condition, with approximately 50 new cases in England and Wales each year, affecting men and women equally with increased incidence with age. Patients' median survival is approximately 6 years, with 50–70% surviving for 5 years and 10–32% for 10 years. Patients most commonly present with acute appendicitis or increasing abdominal girth. Although there are several treatment options, most patients will undergo either standard treatment of debulking surgery or radical surgery and concomitant perioperative intraperitoneal chemotherapy (IPEC) (Sugarbaker procedure).

Objectives

This systematic review examines the clinical and cost-effectiveness of the Sugarbaker procedure for treating PMP and the costs of the procedure in the UK.

Methods

This report was based on a systematic literature review and modelling of costs.

Data sources

The main electronic databases were searched, with English language limits, for the periods up to September 2002. Bibliographies of related papers were assessed for relevant studies and experts contacted for advice and peer review, and to identify additional published and unpublished references.

Study selection

Studies were included if they fulfilled the following criteria, which were applied by one reviewer and checked by a second reviewer, with any disagreements resolved through discussion.

- Intervention: (1) traditional surgery debulking resection of all gross disease, (2) cytoreductive surgery combined with chemotherapy or cytoreductive surgery combined with heated adjuvant IPEC (Sugarbaker procedure).
- Participants: people diagnosed as having PMP characterised by histologically benign tumours with indolent course originating in the appendix.
- Outcomes: survival, recurrence or quality of life as primary outcomes and complications as secondary outcome with a minimum of 2 years' follow-up.
- Design: the highest level of evidence available, which was case series. Economic evaluations were included in the review if they included a comparator (or placebo) and both the costs and consequences (outcomes) or if they were costing studies.

Data extraction

Data extraction and quality assessment were undertaken by one reviewer and checked by a second reviewer, with any disagreements resolved through discussion. The quality of case series was assessed using criteria recommended by the NHS Centre for Reviews and Dissemination (University of York). The quality of economic studies was assessed for their internal validity using a standard checklist, and external validity using a series of relevant questions.

Study synthesis

The clinical effectiveness of the Sugarbaker procedure for PMP was synthesised through a narrative review with full tabulation of results of all included studies. The economic modelling used a Monte-Carlo simulation model, populated UK price data, to estimate likely UK costs.

Results

Number and quality of studies, and direction of evidence

Five retrospective case-series reports assessing the Sugarbaker procedure met the inclusion criteria for the review. No studies comparing the Sugarbaker procedure with standard treatment, or observational studies of standard treatment were included. When judged using standard criteria for assessing methodological quality, the studies were found to be

of poor quality. Patients with different histopathology may have been included in the studies. Details of cytoreductive surgery and chemotherapy differed between studies and not all patients within a series received the same treatment.

Summary of benefits

There appears to be some benefit for people with PMP who undergo treatment with the Sugarbaker procedure. People with PMP have an estimated 5-year and 10-year survival of approximately 50% and 18%, respectively. In contrast, the survival rate of patients following the Sugarbaker procedure is about 90% at 2 years, 60% to about 90% at 3 years, depending on details of IPEC, and 60% to about 68% at 10 years. The percentage of patients with no evidence of disease at the end of follow-up after the Sugarbaker procedure ranged from 41 to 82%. Similarly, the percentage of patients alive with disease at the end of follow-up ranged from 9 to 35%. Mortality due to disease ranged from 2 to 31% in the included studies of the Sugarbaker procedure. Commonly reported complications of the Sugarbaker procedure were anastomotic leaks, fistula formation, wound infection, small bowel perforations/obstructions and pancreatitis.

Costs

No cost-effectiveness or high-quality cost evidence was included in the systematic review. One study of poor methodological quality and set in the USA was found. This study, together with UK unit price data and expert advice, was used to populate a Monte-Carlo simulation model to estimate the marginal cost of operating a service to provide treatment for PMP using the Sugarbaker technique rather than standard treatment. The Monte-Carlo simulation model did not include the costs incurred in setting up the specific service or training the staff. The results of the Monte-Carlo simulation model showed that the cost for one patient over a maximum of 5 years would be about £9700, with a standard deviation of about £1300. The US study showed a ten-fold higher cost. However, the two studies may not be entirely comparable owing to differences in the provision of the specific service and the organisation of the health service.

Cost-utility

No relevant data were available.

Sensitivity analyses

The Monte-Carlo analysis showed that the variation around the mean was not very high. The most likely factor influencing the variation of the costs was the length of procedure. No sensitivity analysis could be done of the alternative treatment.

Conclusions

Limitations of the calculations

The economic results should be seen as merely an example of the likely marginal costs of the Sugarbaker procedure. No policy decision can be made from cost statements without more information about the current alternative. Other questions concerning the capacity and finances of the chosen method have to be left to others.

Implications of Sugarbaker for PMP

If the National Specialist Commissioning Advisory Group were to support the development of additional specialist centres within the NHS, there may be several barriers to implementation. The Sugarbaker procedure requires trained and experienced staff and inevitably there will be the need for a period of training and time costs involved in developing the appropriate teams. Although the procedure requires some specialist equipment and maintenance, such as smoke evacuators, these should have limited effect on setting up the service. PMP is a relatively rare condition with approximately 50 new cases per year in the UK and the impact of an increase in the demand for services should be limited.

Recommendations for research

Evidence is needed for the effectiveness of maximal cytoreductive surgery compared with surgical debulking, using different intraoperative IPEC strategies, and for the effectiveness of treatments in patients who have residual disease following maximal efforts at cytoreduction. Research should take the form of high-quality prospective cohort studies with economic evaluations. Studies should be in histologically homogeneous groups and follow-up should be long enough to assess outcomes such as mortality, survival, recurrence, morbidity, complications and quality of life.

Publication

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