Reviewer’s report

Title: The Cost of Anal Cancer in England: Retrospective hospital data analysis and cohort model

Version: 1

Date: 3 April 2014

Reviewer: Constanza Vargas Parada

Reviewer’s report:

This review has also been attached in case of format problems.

Summary

The main objective of this manuscript was to determine the average cost associated to a rare disease, anal cancer, but whose incidence has increased worldwide becoming itself a threatening health issue. In order to estimate these costs, the authors had to first determine the number of anal cancer cases per year registered in the NHS. Due to lack of data and information the authors developed a Markov model to simulate the treatment pathway of anal cancer and determine the average cost per case. The authors acknowledged data limitations and the corresponding assumptions that had to be made.

Strengths

1. Conflicts of interest were disclosed.
2. The data sources are adequate for the purpose of the study.
3. The limitations of the study are analyzed and discussed by the authors.
4. The manuscript is written in an acceptable way.
5. The results reported in this study can be used for further research as new treatment protocols and preventive strategies become available.
6. For the valuation part of costs, the authors did a separate analysis, with and without inflation using CPI.

Discretionary Revision

7. The retrospective part of this study involved reviewing patient data. It is unclear if the authors required an ethics committee approval.
8. Strong arguments are presented supporting the research. Recent studies on the epidemiology of anal cancer show evidence regarding some possible explanations for the increased incidence of the disease (i.e. Use of HAART, change in sexual behavior, etc.). I would suggest considering this as additional information in the background section.
9. The authors clearly specify the two main objectives of the study which are to estimate the annual costs of treating anal cancer and the cost of treating a single case. However, I would suggest specifying the fact that average costs were
measured (page 5, lines 1 and 2). This is in accordance to the methods described.

10. The methods are divided into two parts. The first aims to determine the number of anal cancer cases. It is unclear if these cases correspond to incident or prevalent cases.

11. The mean start age for males and females reported in the results section (page 9) was not shown in the tables. This could be stated at the end of the sentence (i.e. data not shown).

Minor Essential Revision

12. The study’s main objective was to estimate the costs of anal cancer, however, to do so, the authors had to estimate the number of anal cancer cases diagnosed in England for which a retrospective review was required and performed. I would suggest considering this to explicitly be mentioned in the title of the study (i.e. Incidence and cost of anal cancer in England…). In addition, the whole manuscript talks about a Markov model and never mentions the word cohort. In fact, the word cohort is only mentioned in the title. I would suggest reformulating the title or otherwise explaining how the cohort model is actually defining the methods part of the study.

13. The methods section should better describe the anal cancer cohort considered in this study.

14. The study doesn´t mention the software used for the data analysis.

15. As mentioned by the authors, there is data deficiency and several assumptions had to be considered. All these assumptions bring uncertainty around the final estimate so an uncertainty analysis would be adequate. This way, cost estimates could be reported within a confidence interval.

16. Due to lack of information and data, the authors built a Markov model used to simulate the treatment pathway/natural history of the disease in order to obtain the final average cost. It is unclear for which year the costs are presented.

Major Compulsory Revisions

17. The study question doesn’t specify the perspective from which the study was conducted. This is relevant for the identification of costs as these will depend on the perspective (i.e. societal, healthcare sector, patient, etc.). However, we might infer the authors considered the healthcare sector’s perspective as most cancers are treated and diagnosed at hospitals. In addition, I would also suggest explicitly mentioning the unit of measurement of costs and the year (i.e. 2011 British pounds) to better define the question of this study.

18. As mentioned by the authors, the aim is to estimate the average 10-year cost of treating anal cancer. Costs may need to be adjusted for differential timing when measured. Future costs (i.e. from year 1 to year 10) should be discounted in order to represent the present value. If no discount rate was to be considered in this study, arguments should be stated supporting this decision.

Level of interest: An article of importance in its field
Quality of written English: Acceptable

Statistical review: Yes, but I do not feel adequately qualified to assess the statistics.

Declaration of competing interests:

Have you in the past five years received reimbursements, fees, funding, or salary from an organisation that may in any way gain or lose financially from the publication of this paper, either now or in the future?

From 2009 till 2011 I worked at MSD Chile at the medical department (I received salary). This article may be related to future preventive vaccination against HPV related anal cancer. Since 2012 and at present, no relationship with the pharmaceutical industry.