Cost effectiveness of mass screening for coeliac disease is determined by time-delay to diagnosis and quality of life on a gluten-free diet


Record Status
This is a critical abstract of an economic evaluation that meets the criteria for inclusion on NHS EED. Each abstract contains a brief summary of the methods, the results and conclusions followed by a detailed critical assessment on the reliability of the study and the conclusions drawn.

CRD summary
The objective was to examine the cost-effectiveness of a mass screening programme for coeliac disease in young adults, compared with no screening, and to identify the factors affecting the cost-effectiveness of the programme. The authors concluded that mass screening was cost-effective, but shortening the time from symptom onset to diagnosis, by educating medics, could reduce the value of screening. A conventional cost-effectiveness framework was used and various aspects of uncertainty were considered. The authors’ conclusions appear to be valid.

Type of economic evaluation
Cost-utility analysis

Study objective
The objective was to examine the cost-effectiveness of a mass screening programme for coeliac disease, in young adults, compared with no screening, and to identify the factors affecting the cost-effectiveness of the programme.

Interventions
The screening consisted of the determination of human immunoglobulin A anti tissue transglutaminase antibody levels in the blood. A positive serology was confirmed by an intestinal biopsy. The comparator was no screening, where coeliac disease was diagnosed based on its symptoms.

Location/setting
USA/primary care.

Methods
Analytical approach:
The analysis was based on a state-transition Markov model, with a lifetime horizon. The authors stated that the perspective of the third-party payer was adopted.

Effectiveness data:
A systematic review of the literature was undertaken in the MEDLINE database to identify the relevant sources of data. These included meta-analyses, cohort studies, and retrospective analyses that were published in European countries or the USA. Where multiple sources were found, the authors used their judgement to select the most appropriate estimates for the model. In general, these were biased against screening. The prevalence of coeliac disease was a key input to the model.

Monetary benefit and utility valuations:
The utility values for patients on a gluten-free diet and with the consequences of coeliac disease (irritable bowel syndrome and iron-deficiency anaemia) were from published studies that used the Short Form (SF)-36 Health Survey and converted these scores to utility weights, using a published equation.

Measure of benefit:
Quality-adjusted life-years (QALYs) were the summary benefit measure and they were discounted at an annual rate of 3%.
Cost data:
The economic analysis included the costs of symptomatic coeliac disease, follow-up of coeliac disease, endoscopy, and screening tests. Coeliac disease symptoms included those of both irritable bowel syndrome and iron-deficiency anaemia. The costs were from published literature and Medicare data. They were in US dollars ($) and a 3% annual discount rate was applied. Some costs were at 2003 prices and some were at 2004 prices.

Analysis of uncertainty:
A multivariate sensitivity analysis, based on a Tornado process, was carried out to determine the influence of all variables on the model outcomes. One-way sensitivity analyses were undertaken on the most influential inputs. The ranges for the estimates were from published literature, except for the cost of follow-up of anaemia, which was assumed. A second-order Monte Carlo simulation was carried out, using beta distributions for the clinical inputs and triangular distributions for the economic inputs.

Results
The projected costs were $24.94 with no screening and $158.64 with screening. The corresponding QALYs were 26.9031 and 26.90579. The incremental cost per QALY gained with screening over no screening was $48,960.

The most influential inputs were the time from onset of symptoms to diagnosis of coeliac disease, the utility of treated coeliac disease, and the prevalence of coeliac disease. At a threshold of $50,000 per QALY, screening was not cost-effective with a time to diagnosis of less than 5.9 years (6 years in the base case), or with a utility for treated coeliac disease of less than 0.978 (0.98 in the base case). The acceptability curve showed that the probability of screening being cost-effective was 60%.

Authors’ conclusions
The authors concluded that mass screening was cost-effective, but shortening the time from symptoms appearing to diagnosis, by increasing the awareness of medics, could reduce the value of screening.

CRD commentary
Interventions:
The selection of the comparators was appropriate as the screening strategy was compared against the usual care, in many settings, of no screening.

Effectiveness/benefits:
A valid approach was used to identify the relevant sources of data. The authors stated that the review followed conventional guidelines to find only high-quality data, but very few details of the studies were given and they were conducted in several countries. More details of these studies would have been helpful in judging the validity of the inputs. The key information on the derivation of the utility values was provided. There was a lack of published evidence for the utility of patients with treated coeliac disease. QALYs were an appropriate benefit measure given the impact of the disease on survival and health-related quality of life.

Costs:
The economic analysis was consistent with the perspective of the third-party payer. The authors were based in Israel, but the costs were from the US setting. The sources were reported and were mainly other published studies. Most costs were presented as category totals, reducing the transparency of the analysis. The price year was not explicitly reported. The cost estimates were varied in the sensitivity analysis.

Analysis and results:
The expected costs and QALYs of the two strategies were clearly reported and were appropriately synthesised, using an incremental approach. The uncertainty was satisfactorily assessed, using various methods, and the findings were clearly reported and discussed. The authors stated that the base-case findings were robust, given that their assumptions favoured no screening, but some parameters strongly influenced the results. Conventional discounting was applied to both the costs and benefits.

Concluding remarks:
A conventional cost-effectiveness framework was used and various aspects of uncertainty were considered. The authors' conclusions appear to be valid.

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