Evaluation of the endomysial antibody for celiac disease: operating properties and associated cost implications in clinical practice
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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.

Health technology
Endomysial antibody (EMA) test for the diagnosis of celiac disease.

Type of intervention
Diagnosis.

Economic study type
Cost-effectiveness analysis.

Study population
Patients with suspected celiac disease (gluten-sensitive enteropathy).

Setting
Secondary care. The economic study was conducted in Ontario, Canada.

Dates to which data relate
All EMAs performed in the authors' setting between January 1995 and December 1996 were reviewed. Studies on the same topic published between 1983 and 1997 were also considered. The dates of the resource use data and the prices are unclear.

Source of effectiveness data
Effectiveness data were derived from a single study, literature review and group consensus by the authors.

Link between effectiveness and cost data
Costing was performed using local hospital data and the Ontario Health Insurance Plan Schedule of benefits; 25 patients with celiac disease were surveyed in order to determine rates of medical utilisation and the cost of a gluten-free diet. It is not clear whether these patients were from the same patient sample as that used in the effectiveness analysis.

Study sample
Power calculations were not used to determine sample size. 248 EMAs were reviewed in 159 females and 89 males (age range: 1 to 81 years, mean age 25 years). The 248 EMA results were compared with small bowel biopsy in 66 patients who had undergone both tests.

Study design
This was a retrospective case series carried out in a single centre.

**Analysis of effectiveness**
The main health outcomes used in the analysis were the sensitivity and specificity of EMA as predictor for positive biopsy results.

**Effectiveness results**
EMAs had a sensitivity of 95% and specificity of 64%. The only predictor of positive biopsy that reached statistical significance was a positive EMA.

**Clinical conclusions**
Diagnosis of celiac disease presents a difficult problem for clinicians. Because the patients are frequently children, clinicians and parents are often reluctant to perform small bowel biopsy, a relatively invasive procedure, to make the diagnosis. Serological tests are more acceptable; EMA in particular has high sensitivity and specificity in select populations at risk of having celiac disease.

**Modelling**
Regression analysis was used to look for predictors of positive EMA results and positive biopsy results. A cost-minimisation model from a societal perspective was used to evaluate cost differences across three strategies: EMA for all patients, small bowel biopsy for all patients and EMA followed by biopsy if positive.

**Outcomes assessed in the review**
Probabilities of celiac disease complications with and without gluten free diet (GFD) and of compliance with GFD were derived from the literature.

**Study designs and other criteria for inclusion in the review**
9 studies relating to celiac disease were included in the review, but the main source used was Holmes et al (1989). No particular study designs were reported.

**Sources searched to identify primary studies**
Not stated.

**Criteria used to ensure the validity of primary studies**
Not stated.

**Methods used to judge relevance and validity, and for extracting data**
Not stated.

**Number of primary studies included**
9 studies were included in the review.

**Methods of combining primary studies**
Not combined.
Investigation of differences between primary studies
Not performed.

Results of the review
Probabilities were as follows: complication on GFD - 0, complication no GFD - 0.10, noncompliant GFD - 0.44.

Methods used to derive estimates of effectiveness
Estimates of effectiveness were also based on the authors’ opinions.

Estimates of effectiveness and key assumptions
General consensus was used to determine the probabilities for: symptoms off GFD, celiac positive (0.95), symptoms off GFD, celiac negative (0.75) and symptoms on GFD, celiac positive (0.17).

Measure of benefits used in the economic analysis
The authors did not provide any measure of benefits as they considered all tests to have similar effectiveness.

Direct costs
Direct costs were considered from a societal perspective and discounted at a rate of 5% per year. Costing was performed using local hospital data and the Ontario Health Insurance Plan Schedule of benefits; 25 patients with celiac disease were surveyed in order to determine rates of medical utilisation and the cost of a gluten-free diet. The following costs were estimated: EMA, small bowel biopsy, GFD (lifelong and discounted), delay in diagnosis (GFD + tests), and complications of celiac disease (e.g. lymphoma). Costs/quantities were not reported separately. The price year was not stated.

Statistical analysis of costs
Not applicable.

Currency
US dollars ($).

Sensitivity analysis
All probabilities and cost estimates were subjected to a one-way sensitivity analysis across their plausible ranges to determine whether inaccuracies in these estimates would have a significant impact on the preferred strategy.

Estimated benefits used in the economic analysis
Not applicable.

Cost results
EMA as a diagnostic test for celiac disease was the most expensive strategy, with a cost of $3,174 per patient assessed. The strategy of small bowel biopsy for all patients cost $997, and a strategy of EMA followed by small bowel biopsy for positive patients cost $866 per patient. The results were sensitive to the cost of GFD, the specificity of EMA and the cost of small bowel biopsy.

Synthesis of costs and benefits
Not applicable.

Authors' conclusions
The EMA is best used as a screening test from both a clinical and cost perspective.

CRD COMMENTARY - Selection of comparators
The reason for the choice of the comparators (small bowel biopsy, EMA test and EMA test followed by small bowel biopsy for positive patients) is clear, as all three strategies were used in the authors' setting. You, as a database user, should consider if this applies to your own setting.

Validity of estimate of measure of benefit
It is not clear to what extent all relevant studies were included as there is no evidence for a systematic search of the literature. As all strategies were considered to be of similar effectiveness the authors did not introduce a measure of benefit.

Validity of estimate of costs
The cost methodology is clearly presented and no important cost items appear to have been omitted. A cost-minimization analysis was carried out.

Other issues
Cost data may not be generalisable to other settings or countries.

Source of funding
None stated.

Bibliographic details

PubMedID
9459047

Other publications of related interest

Indexing Status
Subject indexing assigned by NLM

MeSH
Adolescent; Adult; Aged; Aged, 80 and over; Autoantibodies /analysis /economics; Biopsy /economics; Celiac Disease /diagnosis /diet therapy /immunology /pathology; Child; Child, Preschool; Cost Control; Costs and Cost Analysis; Diet, Protein-Restricted; Dietary Proteins /administration & dosage; Evaluation Studies as Topic; Female; Fluorescent Antibody Technique /economics; Forecasting; Glutens /administration & dosage; Humans; Immunoglobulin A /analysis; Infant; Intestine, Small /pathology; Logistic Models; Male; Middle Aged; Multivariate Analysis; Myofibrils /immunology; Probability; Sensitivity and Specificity

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