Systematic Review

Health economic evaluations in orthodontics: a systematic review


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Summary

Background: Economic evaluation is assuming increasing importance as an integral component of health services research.

Aim: To conduct a systematic review of the literature and assess the evidence from studies presenting orthodontic treatment outcomes and the related costs.

Materials/methods: The literature review was conducted in four steps, according to Goodman's model, in order to identify all studies evaluating economic aspects of orthodontic interventions. The search covered the databases Medline, Cinahl, Cochrane, Embase, Google Scholar, National Health Service Economic Evaluation Database, and SCOPUS, for the period from 1966 to September 2014. The inclusion criteria were as follows: randomized controlled trials or controlled clinical trials comparing at least two different orthodontic interventions, evaluation of both economic and orthodontic outcomes, and study populations of all ages. The quality of each included study was assessed as limited, moderate, or high. The overall evidence was assessed according to the GRADE system (The Grading of Recommendations Assessment, Development and Evaluation).

Results: The applied terms for searches yielded 1838 studies, of which 989 were excluded as duplicates. Application of the inclusion and exclusion criteria identified 26 eligible studies for which the full-text versions were retrieved and scrutinized. At the final analysis, eight studies remained. Three studies were based on cost-effectiveness analyses and the other five on cost-minimization analysis. Two of the cost-minimization studies included a societal perspective, i.e. the sum of direct and indirect costs. The aims of most of the studies varied widely and of studies comparing equivalent treatment methods, few were of sufficiently high study quality. Thus, the literature to date provides an inadequate evidence base for economic aspects of orthodontic treatment.

Conclusion: This systematic review disclosed that few orthodontic studies have presented both economic and clinical outcomes. There is currently insufficient evidence available about the health economics of orthodontic interventions. Further investigation is warranted.

Introduction

Economic evaluation has become an integral component of health services research in recent years and is likely to assume increasing importance for decision making in dental services in the future (1). The main reason is that resources within the health sector (personnel, time, facilities, equipment, and knowledge) are limited (2). Thus, failure to analyse economic aspects of dental health services may result in unsustainable over expenditure or...
Economic evaluations involve techniques that were developed in order to support decision making. In health care settings, cost evaluations provide only limited information for this process. In allocating resources, including dental care and orthodontics, health services purchasers need to take into account not only evidence of clinical effectiveness of treatment procedures but also relative costs, i.e. ‘value for money’ (4). Two features characterize health economic evaluation: it concerns the relationship between costs (input) and consequences (output) and preferential selection of diagnostic or therapeutic options, implying alternative ways of allocating resources.

Four main analyses are available for economic evaluations (2):
A **cost-effectiveness analysis** is characterized by analysis of both costs and outcomes, where the magnitude of the outcomes of the various methods might differ. These studies express effectiveness in a single dimension in order to enable direct comparison of costs.
A **cost-minimization analysis**, which is a type of cost-effectiveness analysis, applicable when two or more treatment methods achieve the same outcomes (e.g. correction of crossbite) and the aim is to identify the least expensive method.
A **cost-utility analysis**, focusing on both costs and quality of the health outcome produced by the treatments: used, for example, in health-related quality-of-life studies.
A **cost-benefit analysis**, which is characterized by the fact that health outcome is also expressed in monetary units. This kind of analysis is seldom used but has been implemented, for example, when evaluating vaccination programs.

Economic evaluations with a societal perspective include calculations of direct and indirect costs. Direct costs are those directly associated with diagnostic or treatment interventions, i.e. clinical interventions, costs of material, costs for the premises and equipment, maintenance, cleaning, and staff costs. Indirect costs arise as consequences of absence from work for treatment, often defined as corresponding to loss of income incurred by the patient, or the patient’s parents, in taking time off from work for clinical appointments and travel to and from the clinic. The societal costs are defined as the sum of direct and indirect costs.

Delivery of equitable health care requires assessment of the economic implications of different interventions (1). In orthodontics, economic analysis is also important as a basis for decision making in planning and management of dental care.

The aim of this study was to conduct a systematic review of published studies of health economics in orthodontics, assessing the evidence from studies presenting an analysis of costs associated with the treatment outcomes.

### Materials and methods

The literature review was conducted in four steps, according to Goodman’s model (5).

#### Definition of the research question

The question to be addressed was ‘What evidence is available from studies combining costs and orthodontic treatment outcomes?’

#### Formulation of a plan for the literature search

A literature search was conducted to identify all studies evaluating economic aspects of orthodontic interventions. The period to be covered was from 1966 to September 2014. The following databases were searched: Medline, Cinahl, Cochrane, Embase, Google Scholar, National Health Service Economic Evaluation Database, and SCOPUS.

The computerized search was conducted with the assistance of a specialist in informatics at the Library, Faculty of Odontology, Malmö University, Sweden. The following search syntax was used:


#### Literature search and retrieval of publications

At the start of the review, consensus was reached on the criteria presented in Table 1.

The potential eligibility of studies was determined independently by five researchers. The titles and abstracts of all potentially relevant studies were independently reviewed. The full-text version of an article was retrieved if at least one of the five reviewers considered the study to be relevant or if the title and abstract did not provide sufficient information. Using a preset protocol and the initial inclusion and exclusion criteria, the full-text versions of the articles were independently analysed and evaluated by the five researchers. In cases of inter-examiner disagreement, each article was reread and discussed until consensus was reached. The reference lists of articles deemed eligible were also hand-searched for additional studies.

#### Data extraction, quality assessment, interpretation, and evaluation of evidence

Data from studies were tabulated as follows: Reference, country, aim of the study, study design, study population, economic and orthodontic outcomes, and finally, study quality (Table 2).

The quality assessment was performed according to protocols independently by the researchers and disagreement over assessment was resolved by discussion to achieve consensus. Study quality was denoted as limited, moderate, or high, according to the checklist proposed by Drummond et al. (2). The strength of the scientific evidence based on study quality from included studies was undertaken according to the GRADE system (14).

#### Results

The PRISMA-compliant selection process (15) is presented in Figure 1. Initially, 1838 articles were identified, of which 989 were deleted as duplicates. After application of the inclusion and exclusion criteria, the full-text versions of 26 studies remained (Figure 1).
Table 1. Inclusion and exclusion criteria used in this systematic review.

<table>
<thead>
<tr>
<th>Inclusion criteria</th>
<th>Exclusion criteria</th>
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<tbody>
<tr>
<td>Studies comparing at least two different orthodontic interventions, including evaluation of economic and orthodontic outcomes.</td>
<td>Incomplete economic analysis</td>
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<td>Randomized controlled trials (RCTs) or controlled clinical trials (CCTs)</td>
<td>Practice economic evaluations</td>
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<tr>
<td>Study populations of all ages</td>
<td>Case reports and case series</td>
</tr>
<tr>
<td>Full-text articles written in all languages</td>
<td>Descriptive articles, opinions, editorials and reviews</td>
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</table>

A further 18 of these studies were excluded for various reasons (Figure 1). Eight studies then remained for final assessment, four from Sweden (6–8,13) and two each from the Netherlands (11,12) and UK (9,10). Of these studies, there were four on different orthodontic appliances (7–9,13), two on retainers (6,10), one on infant orthopaedic treatment (11), and finally, one study on cleft palate treatment (12). The studies by Konst et al. (11) and Severens et al. (12) concerned the same cohort from the Dutch-cleft study (16), with cost data based on resource utilization from the latter randomized controlled trial (RCT) (16). Of the four studies on appliances, only two included comparable forms of appliance treatment, namely crossbite treatment using Quad Helix or expansion plates (7,13).

Five studies were based on cost-minimization analyses (6–9,13), although in the study by Hermanson et al. (13), the cost-minimization analysis was erroneously described as a cost–benefit analysis. Three studies used a cost-effectiveness analysis (10–12). Only two studies (6,7) included a societal perspective.

Evaluation of study quality and evidence
The quality of four (8,11–13) of the eight studies was assessed as moderate, three (6,7,9) as moderate, and one as high (10). The reason for assessing the articles from Konst et al. (11) and Severens et al. (12) as limited were that the number of included patients did not correspond to the number of patients included in the original RCT. Thus, in the Dutch-cleft study by Prahl et al. (16), which formed the basis for the calculations, 23 patients per group was estimated to be needed. However, in the study by Severens et al. (12), 23 and 20 patients per group were used for the economic evaluation. The other two studies (8,13) of limited study quality both had a retrospective design. One RCT from UK (10), presented with high transparency and included almost 400 patients, was assessed as high study quality.

Under the GRADE system for grading the evidence, study quality is denoted as high, moderate, or limited (Table 2). Because of the broad range of aims among most of the studies (6,8–12), and the few studies of adequate quality covering the same comparison of appliances (7,13), no evidence base for economic aspects of orthodontic treatment emerged.

Discussion
No evidence of cost effectiveness of orthodontic interventions emerged from this review, primarily because so few relevant studies could be included and all but two concerned different methods. Of the two studies investigating the same orthodontic methods (7,13), only one (7) was assessed as having moderate study quality. However, when there are few relevant published studies, as in the present case, it is important to bear in mind that lack of evidence does not necessarily imply lack of effect, including economic outcome. Thus, there is an urgent need for further economic analysis of orthodontic interventions. It can also be noted that our results coincide with a recent systematic review regarding results for economic evaluation of diagnostic methods used in dentistry (17). That review highlighted a paucity of economic evaluations of diagnostic methods and a need for studies from various perspectives with well-defined research questions and measures of the cost and effectiveness (17).

Of the eight studies remained for the final analysis, four studies considered different orthodontic appliances (7–9,13) and of these only two included comparable forms of appliance treatment, namely crossbite treatment using Quad Helix or expansion plates (7,13). In addition, two studies concerned retainers (6,10), one on infant orthopaedic treatment (11) and finally one study on cleft palate treatment (12). Since few studies have been undertaken and methodologically there was between-study heterogeneity, no cumulative evidence or meta-analysis could be assessed regarding the health economics of orthodontic interventions. Consequently, much of what we practice lacks sufficient evidence about the health economics of orthodontic interventions.

The studies assessed in this review were predominantly from Europe and especially from Sweden. Thus, there were few or no studies from other parts of the world. It seems that Swedish researchers have taken the initiative in presenting combined economic and orthodontic outcomes in studies of health economics. This may be seen in the context of the Swedish system of subsidized dental care, including publicly funded orthodontic treatment for children and adolescents.

The question has arisen as to whether lack of evidence based on economic evaluations of orthodontic interventions presents a problem. Some authors, for instance Anderson (18), claim that systematic reviews of economic studies are of limited relevance, primarily because of problems of transferability: as the costs incurred in studies from one country are seldom representative of another country (18). Thus, studies of cost effectiveness are seldom relevant for other countries and an example of this may partly be mirrored in a Finnish study (19) where the factors affecting the costs of children’s free orthodontic treatment were studied and significant differences in treatment patterns and costs between different health centres were found.

What could be done to enhance comparisons between economic studies of orthodontic interventions undertaken in different countries? One way would be to include specific data not only on economic results but also on resource utilization. For instance, specifications of time input by dentists and dental nurses, respectively, and specifications of how overhead costs were calculated, as a percentage or as absolute added costs. Studies from different countries could then be compared more easily. Despite the issue of transferability, assessment of studies identified in systematic reviews is important, as the evidence may serve as guidelines for planning and management of orthodontic interventions.

One attempt to find ways of comparisons between studies of cost effectiveness in orthodontic care is described in a study of exploratory nature (20). There were large differences of cost effectiveness found between included practitioners from seven countries in Europe. However, the differences were mainly due to differences in...
### Table 2. Summary of studies included in the quality assessment, listed in descending order, from the latest publication. RCT = randomized controlled trial.

<table>
<thead>
<tr>
<th>Reference, country</th>
<th>Aim of the study</th>
<th>Study design follow-up</th>
<th>Study population</th>
<th>Economic and orthodontic outcomes</th>
<th>Study quality</th>
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<tbody>
<tr>
<td>Tynelius Edman et al. (6), Sweden</td>
<td>Compare costs for three retention methods; Vacuum formed retainer + canine to canine retainer (V-CTC), vacuum formed retainer + stripping (V-S), and positioner (P)</td>
<td>Costs based piggy-back on one earlier published RCT. 2 years follow-up</td>
<td>Adolescents 17–19 years of age. Group V-CTC: N = 24 (17 girls and 7 boys), 1 dropout. Group V-S: N = 23 (14 girls and 9 boys), 2 dropouts. Group P: N = 22 (13 girls and 9 boys). 3 dropouts</td>
<td>Cost-minimization analysis. Direct, indirect, and societal costs. Equally efficient in retaining orthodontic treatment results. Costs in Euro per patient: V-CTC 457; V-S 451; Positioner 429; V-CTC retainer was the least cost effective</td>
<td>Estimated treatment times. Average times used for calculations. Societal perspective. Intention to treat. Low attrition. Moderate study quality</td>
</tr>
<tr>
<td>Petén et al. (7), Sweden</td>
<td>Compare costs of unilateral posterior crossbite correction using Quad Helix (QH) or expansion plate (EP)</td>
<td>Costs based piggy-back on one earlier published RCT. 3 years follow-up</td>
<td>Group QH: N = 20, mean age 9.0 years (range 7.8–10.2 years), 11 girls and 9 boys. Group EP: N = 20, mean age 8.5 years (range 7.5–9.5 years), 13 girls and 7 boys</td>
<td>Cost-minimization analysis. Direct, indirect, and societal costs. Success rate of correction, transverse expansion. QH and EP had equally successful crossbite corrections. QH offers significant economic benefits over EP. QH had lower direct and indirect costs, also fewer failures needing retreatment. Costs in Euro QH 1031 versus EP 1395 (P &lt; 0.001)</td>
<td>Unclear about variability of some costs compared to treatment time. Societal perspective. Intention to treat analysis. No attrition. Moderate study quality</td>
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<tr>
<td>Laag and Ström (8), Sweden</td>
<td>Compare results and costs at a general dentist clinic (GDC) and a special orthodontic clinic (SOC) for treatment of crowding, increased overjet, lateral crossbite, and dental displacement</td>
<td>Retrospective CCT. Cases at GDC and controls at SOC. Follow-up unknown</td>
<td>Removable appliance at GDC and fixed appliance at SOC. Group GDC: N = 31, mean age 11.4 years (range 8.2–14.2 years), 20 girls and 11 boys. Group SOC: N = 19, mean age 13.1 years (11.1–14.2 years), 16 girls and 3 boys</td>
<td>Cost-minimization analysis. Weighted Peer Assessment Rating (WPAR-index). GDC 69.0 (±10.6) versus SOC 84.1 (±7.1) (P &lt; 0.001). Costs SEK GDC 3.525 versus SOC 5.510. Trained general dentists may achieve sufficiently qualitative results at a reasonably low cost. The results tend to be better at the SOC but more expensive than at the GDC.</td>
<td>Simplified calculations of costs. Retrospective CCT. Non-cooperative patients were not registered, thus only successful treatment was counted. Limited study quality</td>
</tr>
<tr>
<td>O’Brien et al. (9), UK</td>
<td>Evaluate the effectiveness and costs of early versus adolescent orthodontic treatment of Class II division 1 malocclusion</td>
<td>Multicentre. RCT. 10 years follow-up</td>
<td>Group early Twin-block appliance: N = 89, mean age at start 9.7 years (SD 0.98), 41 girls and 48 boys. Removable or fixed appliance for the adolescent group: N = 85, mean age 9.8 years (SD 0.98), 39 girls and 46 boys. Early treatment group: mean age 12.4 years (SD 1.16) at second treatment phase. Adolescent treatment group: mean age 12.1 years (SD 1.1)</td>
<td>Cost-minimization analysis. No difference with respect to skeletal pattern, extraction rate, patient self-esteem. Early twin-block treatment incurred higher costs 3913 versus 3018 $/patient. Early treatment with removable appliance followed by further treatment in adolescence at the appropriate time does not result in any meaningful long-term benefit over one course of treatment started in the late mixed dentition</td>
<td>No sensitivity analysis. Intention to treat analysis. 19% dropout rate. Moderate study quality</td>
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Table 2. Continued

<table>
<thead>
<tr>
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<tr>
<td>Hichens et al. (10), UK</td>
<td>Compare cost-effectiveness of Hawley retainers (HR) and vacuum-formed retainers (VFR)</td>
<td>RCT. 6 months follow-up</td>
<td>Group HR: N = 196, mean age 14.8 years, 124 girls and 92 boys. Group VFR: N = 201, mean age 15.0 years, 119 girls and 82 boys</td>
<td>Cost-effectiveness analysis. VFR was more cost effective than HR from all perspectives, i.e. National Health Service, the practices and the patients</td>
<td>Well performed and good transparency of calculations. Power analysis. Drop-outs presented. High study quality</td>
</tr>
<tr>
<td>Konst et al. (11), The Netherlands</td>
<td>Investigate the cost-effectiveness of infant orthopaedic treatment (IO) of unilateral cleft lip and palate compared with no such treatment (NT) on speech development</td>
<td>Three-centre RCT. 2 years follow-up</td>
<td>20 patients randomized to two groups. Group IO: N = 10, age 2.5 years, 2 girls and 8 boys. Group NT: N = 10, age 2.5 years, 1 girl and 9 boys</td>
<td>Cost-effectiveness analysis. IO: Cost of treatment by orthodontists per patient €1460. NT: €419. ICER: incremental cost-effectiveness ratio, €1041 per 1.34 point speech quality improvement. Regarding speech in cleft patients, IO group had a statistically significant higher rating than the NT group</td>
<td>Selection of subjects unclear, i.e. unknown if consecutive patients. High attrition rate. Interpretation unclear. Number of patients not corresponding to the power analysis in original study. Limited study quality</td>
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<tr>
<td>Severens et al. (12), The Netherlands</td>
<td>Investigate cost effectiveness of complete unilateral cleft palate treatment from pre-surgical orthopaedic treatment (PSOT)</td>
<td>Three-centre RCT. PSOT compared with none-PSOT. 18 weeks follow-up</td>
<td>Group PSOT: N = 23, age 2 weeks, gender distribution not presented. Group none-PSOT: N = 20, age 2 weeks, gender distribution not presented</td>
<td>Cost-effectiveness analysis. Surgical lip closure, no significant difference. PSOT US$ 852 versus none-PSOT US$ 304. Regarding the incremental cost-effectiveness ratio, PSOT was more expensive than none-PSOT and had an insignificantly lower effect</td>
<td>No information about attrition. Number of patients not corresponding to the power analysis in original study. Limited study quality</td>
</tr>
<tr>
<td>Hermanson et al. (13), Sweden</td>
<td>Compare effects, complications, discomfort, and costs of unilateral crossbite correction using Quad-helix appliance (QH) and removable plate appliance (RP)</td>
<td>Retrospective CCT. 2 years follow-up</td>
<td>Group QH: N = 27, mean age 9.8 years (range 7.3–13.0 years), 14 girls and 13 boys. Group RP: N = 17, mean age 9.5 years (range 6.8–14.3 years), 12 girls and 5 boys</td>
<td>Cost-minimization analysis. Successful treatment: QH 26 of 27 and RP 16 of 17. Complications equal. Costs QH: 1223 SEK versus RP 1714 SEK. The cost analysis showed a 40% higher treatment cost with RP than QH. Treatment with RP takes longer than QH treatment</td>
<td>Selection of subjects unclear, i.e. unknown if consecutive patients. Economic analysis not a cost–benefit analysis but cost minimization,thus the authors have labelled the economic analysis incorrectly. Retrospective CCT. Limited study quality</td>
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</table>

Costs, not in effects. The authors stressed the importance of including cost-effectiveness analyses in orthodontics in order to achieve ‘value for money’ (20). Moreover, a retrospective multicentre study from the UK had a focus on costs of direct health services. Costs were assessed of combined orthodontic and surgical treatment of dentofacial deformity. It was demonstrated that orthodontic costs on average comprised 25 per cent of the total treatment cost and this is stated to be inexpensive. Unfortunately, the effectiveness of orthodontic care was not considered in this study (21).

This systematic literature review was conducted according to Goodman’s model (5) and comprised four steps: 1. definition of research question, 2. formulation of a plan for literature search, 3. literature search and retrieval of publications, and 4. data extraction, interpretation, and evaluation of evidence from the literature retrieved. This model fulfills the criteria of repeatability and minimizes the risk that the conclusions will be affected by chance or arbitrariness. In a further step, the study quality of the articles was classified as low, moderate, or high according to criteria suggested by Drummond et al. (2). In order to avoid subjectivity in the assessments, the independent observers evaluated the quality of separate studies and the overall level of evidence. In 90 per cent of cases, the different reviewers’ evaluation and grading of the full-text articles...
were in agreement; consensus on the remainder was reached after rereading and discussion.

One challenge encountered in this systematic review was that although the literature search identified as many as 1838 studies, 989 were deleted as duplicates. Furthermore, 823 articles did not meet the stated objective of the review and only 26 articles remained for the final analysis. This is not, however, an unusual outcome for systematic reviews: the initial literature search was intentionally designed to be broad enough to ensure that no article would be over-looked or disregarded.

Conclusions

This systematic review disclosed that few orthodontic studies have presented both economic and clinical outcomes. There is currently insufficient evidence available about the health economics of orthodontic interventions. Further investigation is warranted.

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References


