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Cost-Effectiveness Analysis of Osteosarcoma Treatment: Insights from a Systematic Literature Review

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INTRODUCTION

- Osteosarcoma is the most common primary malignant bone tumour that arises from mesenchymal cells. It is characterized by areas of uncontrolled or abnormal bone growth and primarily affects adolescents and young adults
- The incidence of osteosarcoma is very rare compared with other cancers, and it is considered an 'ultra orphan' disease.[1] Global annual incidence rates range from 1.5 to 5 cases per million in men and from 2 to 4 cases per million in women[2]
- Standard treatment protocols typically include multi-agent chemotherapy, surgical resection and radiation therapy
- This study emphasizes the necessity for cost-effective treatments to improve patient outcomes and manage healthcare expenses

OBJECTIVES

- This systematic review evaluated model-based economic assessments, specifically cost-effectiveness and cost–utility studies, for osteosarcoma treatments.

METHODS

- Embase® and MEDLINE® were systematically searched through Embase.com, following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines, for English-language studies on the economic analysis of osteosarcoma from 2013 to 2024. Two independent reviewers screened titles and abstracts, with full-text assessments performed by the same reviewers and reconciled by a third reviewer if needed. The study methodology is presented in Table 1

Table 1. SLR methodology

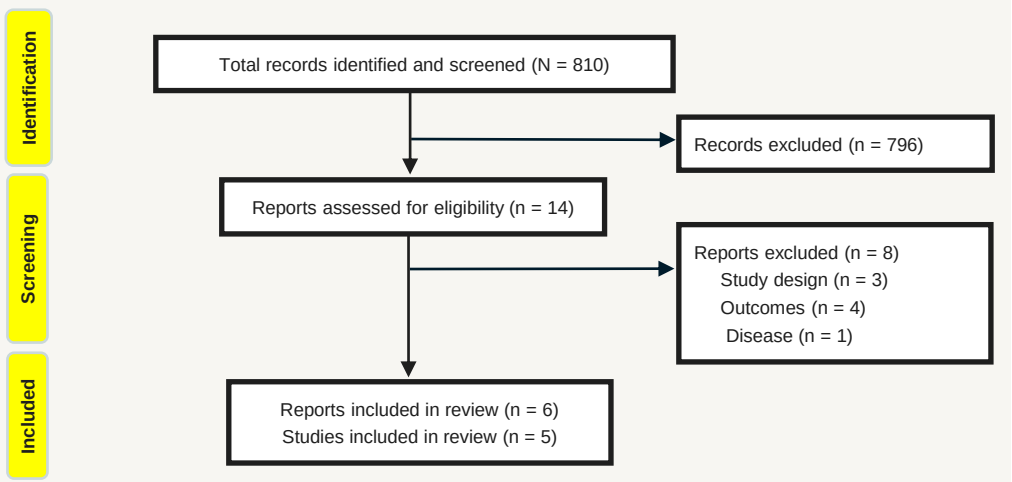
Population	Patients with osteosarcoma
Intervention	No intervention limits
Outcomes	QALYs, LYs, ICER, total costs, indirect and direct cost components
Study designs	Cost-effectiveness analysis Cost-minimization analysis Cost–utility analysis SLR and meta-analysis (for cross referring only)
Country	No geographical limits
Language	English only
Timeline	2013 to 2024

Key: ICER, incremental cost-effectiveness ratio; LY, life year; QALY, quality-adjusted life year; SLR, systematic literature review.

RESULTS

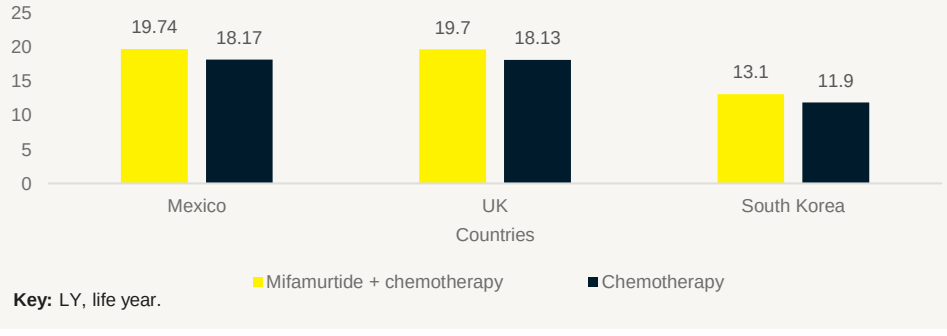
- Out of 810 studies screened, five studies (six reports) focussing on cost-effectiveness analysis for the combination of mifamurtide and chemotherapy were included (Figure 1)
- Among the included studies, two were conducted in South Korea and one each was conducted in Mexico, Spain and the UK
- A Markov model was used for the cost-effectiveness analysis in all the included studies
- Perspectives included the Spanish National Health Service, Mexican public health institutions and the UK National Health Service (NHS)

Figure 1. PRISMA diagram



- Improved life years (LYs) of 19.74, 19.7 and 13.1 were observed when mifamurtide was combined with the chemotherapy regimen for treating osteosarcoma in Mexico, the UK and South Korea, respectively, compared with chemotherapy alone.[1, 3, 6, 7] This combination resulted in an increase of 1.57 LYs in Mexico and the UK, and an increase of 1.2 LYs in South Korea[1, 4, 6, 7] (Figure 2)
- Introducing mifamurtide treatment along with chemotherapy regimens in patients with osteosarcoma led to improved quality-adjusted life years (QALYs) across the countries
- Spain demonstrated the most substantial incremental gain of 3.04 QALYs followed by Mexico, the UK and South Korea with incremental gains of 1.6, 1.34 and 0.96 QALYs, respectively for the combination[1, 4-7] (Figure 3)

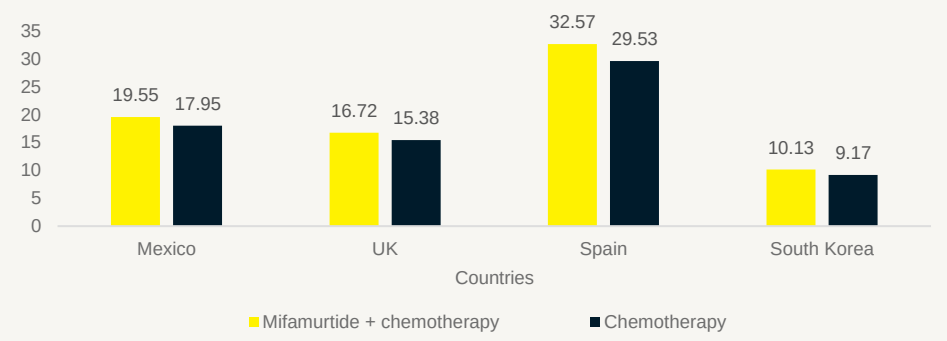
Figure 2. Country-specific LYs



Key: LY, life year.

- Including mifamurtide in combination therapy imposed a significant financial burden due to its considerably higher cost compared with chemotherapy alone (Table 2)
- The incremental cost-effectiveness ratio (ICER) was significant, reflecting the additional costs for improved LYs or QALYs with the combination therapy (Table 3)

Figure 3. Country-specific QALYs



Key: QALY, quality-adjusted life year.

Table 2. Total costs in Mexico, the UK, Spain

Country	Total costs	
	Mifamurtide + chemotherapy	Chemotherapy alone
Mexico	USD 102,635.8	USD 13,148.9
UK	GBP 123,852	GBP 31,481
Spain	EUR 124,201	EUR 22,201

Table 3. ICER values in Mexico, the UK, Spain

Country	ICER Mifamurtide + Chemotherapy vs Chemotherapy alone	
	Per LYs	Per QALYs
Mexico	USD 56,746	USD 55,837
UK	GBP 58,737	GBP 68,734
Spain	-	EUR 76,620

Key: ICER, incremental cost-effectiveness ratio; LY, life year; QALY, quality-adjusted life year.

CONCLUSIONS

- This review of economic studies suggest that mifamurtide offers potential long-term survival benefits for patients with osteosarcoma
- Despite its high initial costs, mifamurtide's cost-effectiveness is justified by its potential to improve long-term survival outcomes
- The cost-effectiveness of mifamurtide varies across different regions and healthcare systems, indicating the need for region-specific economic evaluations
- Despite the benefits of mifamurtide, unmet needs remain for treating osteosarcoma, highlighting the necessity for continued research and development in this area

LIMITATIONS

- None of the studies reviewed provided information on the indirect costs related to osteosarcoma
- While other treatment options exist, this analysis was ultimately limited to examining mifamurtide in combination with chemotherapy. The lack of substantial evidence for alternative treatments further limits the generalizability of the study's findings
- The study was constrained by the limited availability of evidence, which subsequently restricted the geographical regions included in the analysis

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