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The Use of Formal Expert Opinion Elicitation to Inform Model Structure and Inputs in Rare Diseases

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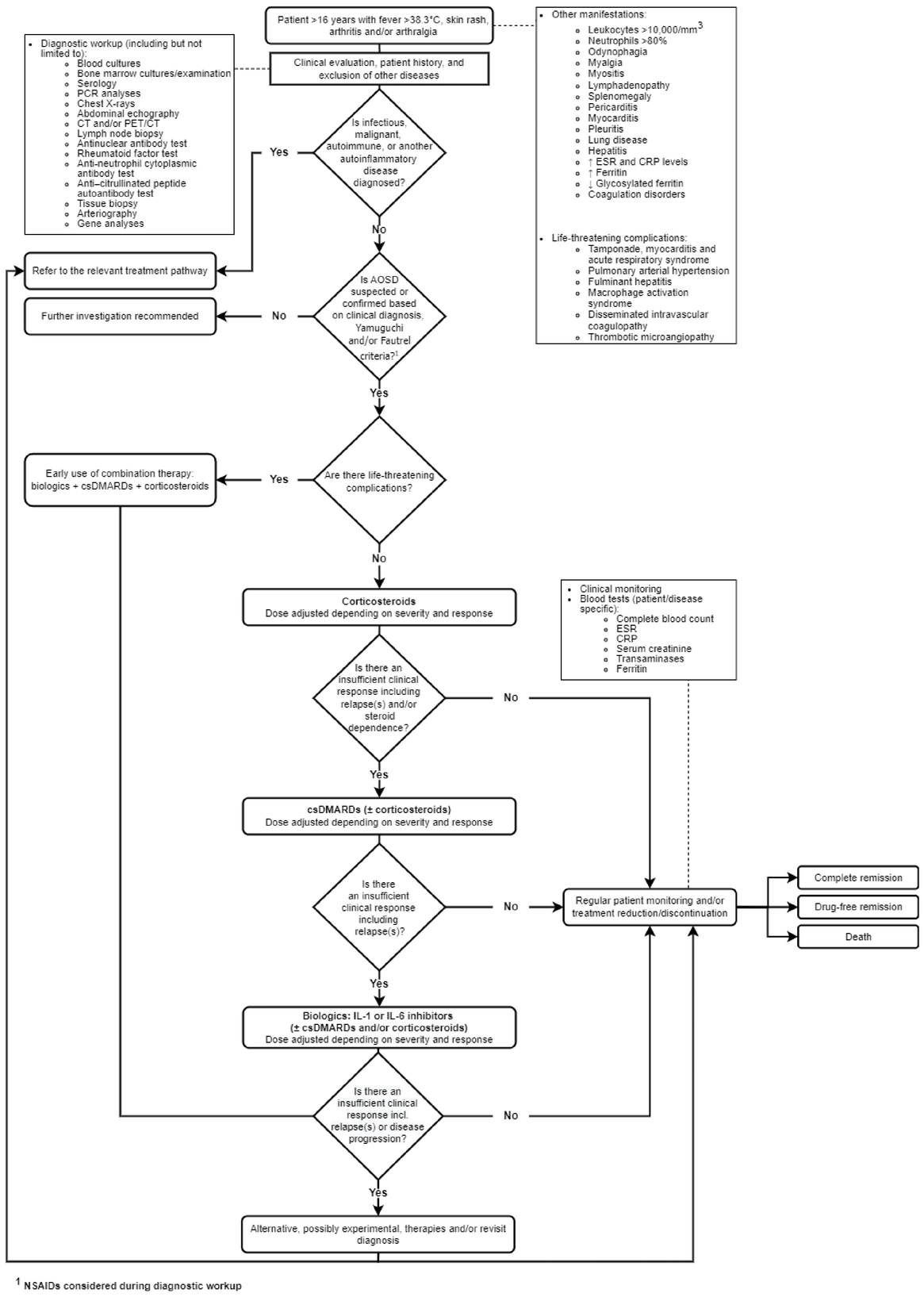
BACKGROUND AND OBJECTIVES

An economic model is composed of two main elements: the model structure, typically identified by investigation of the care pathway, and model inputs, extracted from quantitative data. High uncertainty in both elements could confer a high risk that the economic model mispresents the true cost-effectiveness of the new intervention.

High uncertainty is particularly prevalent in rare disease areas, when the diagnostic pathway is usually complex and variable due to a lack of clinical guidelines, and/or accurate, cheap or non-invasive diagnostic tests to identify the population of interest. Clinical trials for these conditions are also expensive because of the complexity of patient recruitment, leading to a lack of robust evidence of treatment outcomes.

This research developed a robust approach to inform the implementation of a cost-effectiveness model in adult-onset Still's disease (AOSD), a rare systemic inflammatory disorder.

Figure 1: Care pathway for patients with AOSD



[1] NSAIDs considered during diagnostic workup

METHODS AND RESULTS

Figure 2: Methodology used to produce the model structure and inputs for economic analysis of AOSD

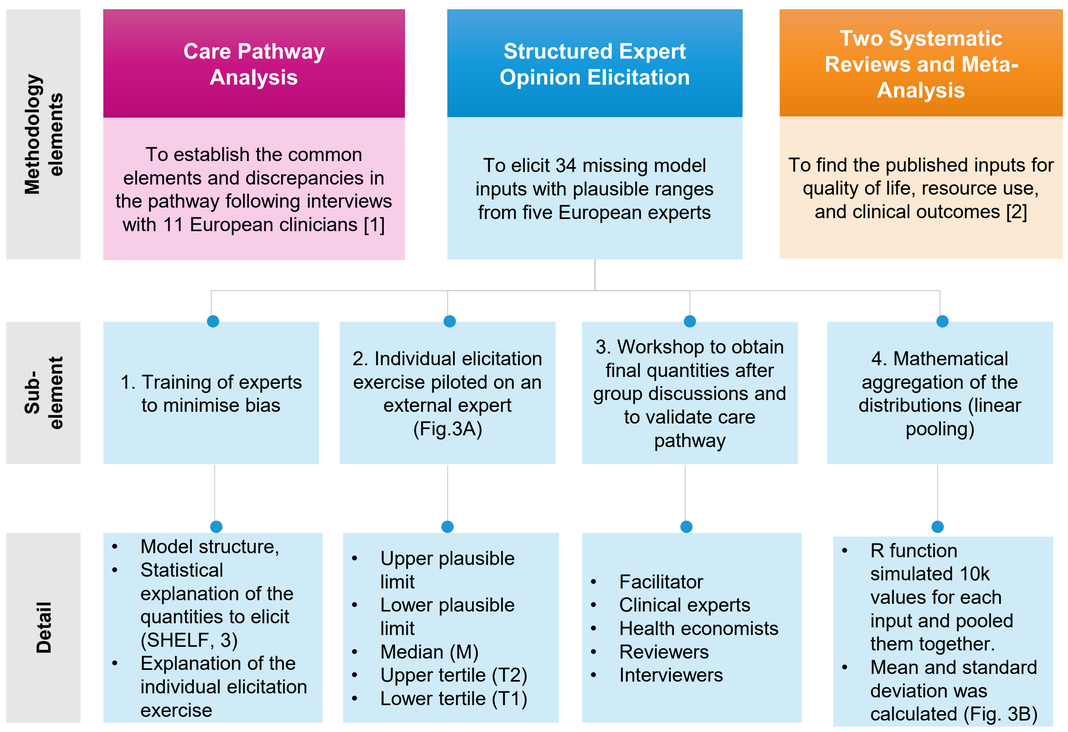
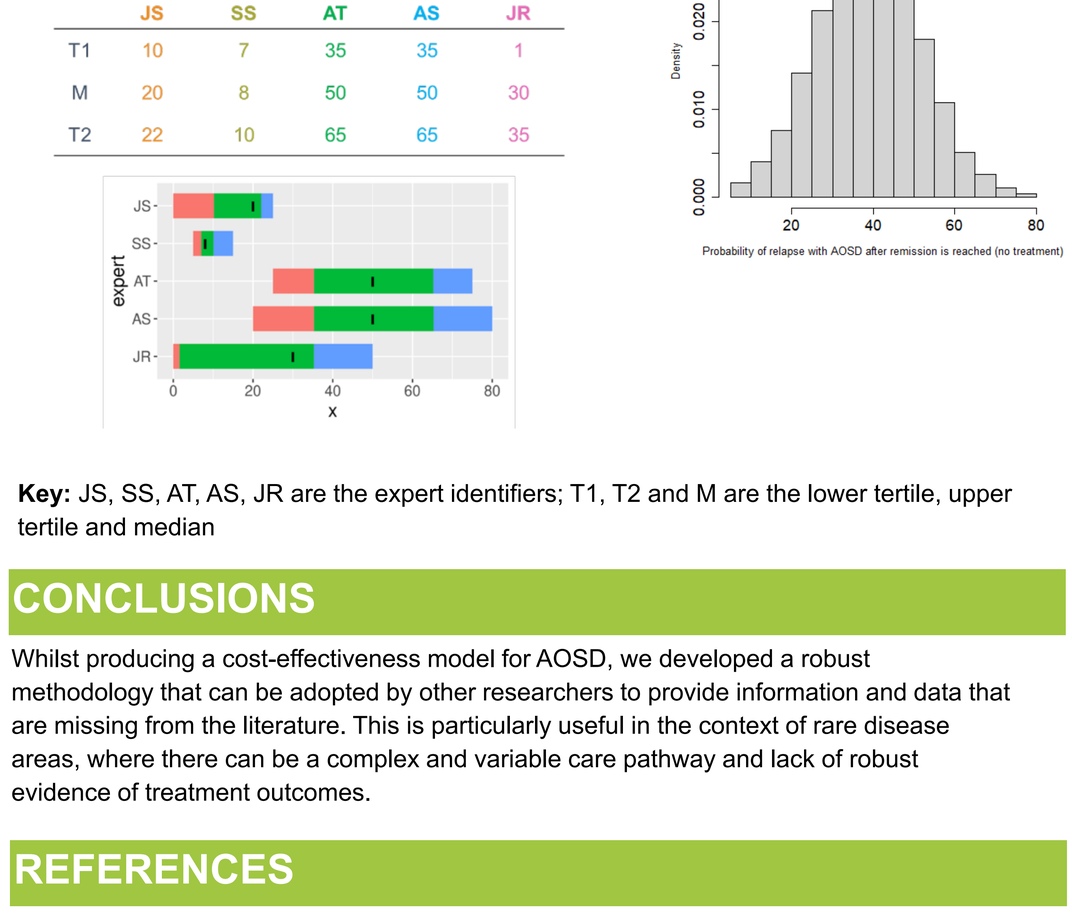


Figure 3: Quantities from the individual elicitation presented at the workshop (A) and the final distribution (B)



Key: JS, SS, AT, AS, JR are the expert identifiers; T1, T2 and M are the lower tertile, upper tertile and median

CONCLUSIONS

Whilst producing a cost-effectiveness model for AOSD, we developed a robust methodology that can be adopted by other researchers to provide information and data that are missing from the literature. This is particularly useful in the context of rare disease areas, where there can be a complex and variable care pathway and lack of robust evidence of treatment outcomes.

REFERENCES

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