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[Challenges and proposed framework for formative research to inform systematic intervention development in rare and unstudied conditions: The case example of Xeroderma Pigmentosum.](#)
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Purpose: To outline the challenges of applying existing systematic intervention development approaches in rare diseases, and to propose a novel framework within which these challenges can be met.

Background: A gap in the provision of self-management and psychosocial interventions to change behaviour and improve health in rare diseases exists, partly due to the difficulty of conducting formative research in such conditions. Challenges include heterogeneity within already small sample sizes, patient burden, and the absence of prior research to guide decision-making. XP is a very rare inherited disease (~100 UK patients), involving an inability to repair ultraviolet radiation (UVR)-induced damage and increased melanoma risk; the only treatment is complete photoprotection. No research in XP has been conducted outside of the genetic literature.

Methods: Using XP and improved photoprotection as a case example, we highlight the necessity of departing from the steps outlined by three intervention development approaches (intervention mapping, UK MRC guidelines for complex interventions, behaviour change wheel), and outline a framework that can be applied to the conduct of formative research in rare diseases. The framework focuses on the sequential or parallel use of mixed-methods (e.g., n-of-1, interviews, reviews of comparable conditions) and the triangulation of gathered data, and provides solutions to challenges including patient burden and the inability to pre-test study materials or intervention content in members of the target population.

Conclusions: The proposed framework offers an alternative that may overcome the limitations associated with intervention development in rare diseases, which will hopefully encourage much-needed work in this field.

250 words