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Background: Patients with Ewing sarcoma or osteosarcoma have a median overall survival of less than 12 months after diagnosis, and a standard treatment strategy has not yet been established. Pharmacological inhibition of MET signalling and aberrant angiogenesis has shown promising results in several preclinical models of Ewing sarcoma and osteosarcoma. We aimed to investigate the activity of cabozantinib, an inhibitor of MET and VEGFR2, in patients with advanced Ewing sarcoma and osteosarcoma. Methods: We did a multicentre, single-arm, two-stage, phase 2 trial in patients with advanced Ewing sarcoma or osteosarcoma recruited from ten centres in the French Sarcoma Group. Key eligibility criteria were aged 12 years or older, Eastern Cooperative Oncology Group performance status of 0-1, and documented disease progression (according to Response Evaluation Criteria in Solid Tumors version 1.1) before study entry. The number of previous lines of treatment was not limited. Patients received cabozantinib (adults 60 mg, children [

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