A 24-year-old man with Von-Hippel-Lindau syndrome and central nervous system (CNS) hemangioblastomas was found to havemultiple leaking retinal capillary hemangioblastomas in the superotemporal periphery of the left eye (A). Systemic treatment was initiatedwith Belzutifan, a hypoxia-inducible factor-2a inhibitor. There was marked improvement in the engorgement and tortuosity of the feedingarterioles and draining veins, and a reduction in the number and size of the retinal capillary and CNS hemangioblastomas after 6 months oftreatment (B)