The aim of the present study was to analyze the prevalence and characteristics of outer retinal tubulation (ORT) by spectral-domain optical coherence tomography (SD-OCT) in patients with macular dystrophies. We performed a retrospective review of the ophthalmic charts of 61 eyes of 32 patients with macular dystrophies. Quality SD-OCT images were analyzed in order to assess the presence and location of ORT, and the integrity of the external limiting membrane (ELM), the photoreceptors inner segment-outer segment junction (IS/OS), and the cone outer segment tips (COST). Overall, ORT was evidenced in 13 eyes (21.3%). When considering only cases with outer retinal disruption, ORT was present in 50% of cases with ELM disruption, 34.2% of cases with IS/OS disruption, and in 24.5% of cases with COST disruption. Only cases with atrophic outer retinal changes in Stargardt’s disease, X-linked retinoschisis and cone dystrophy, showed ORT in the SD-OCT. In conclusion, ORT may be a frequent SD-OCT finding in macular dystrophies with severe disruption of the outer retinal architecture. We hypothesize that ORT correspond to the retinal structures defined histopathologically as rosettes. ORT might be a later event of retinal degeneration, strongly related to the disruption of the ELM.