Protein O-Mannosylation: News and Views

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Protein O-mannosylation is an essential modification among fungi and mammals. It is initiated at the endoplasmic reticulum by a conserved family of protein O-mannosyltransferases (PMT/POMTs). Mutations in human *POMTs* cause alpha-dystroglycanopathies covering a wide spectrum of clinical severities ranging from Walker-Warburg syndrome to limb-girdle muscular dystrophy. Recent work addressing the functional characterization of POMTs and the role of O-mannosyl glycans during mouse embryo development will be discussed.