Introduction: Butane is a hallucinogenic recreational agent often misused by young substance abusers. Butane intoxication is known to cause injury to the nervous system. Previous studies report cases of encephalopathy and coma following intoxication. We present a case of butane intoxication presenting with symptoms suggestive of Wernicke’s encephalopathy. Case Report: A young man with multiple substance abuse history was admitted because of sudden onset of diplopia and gait instability. Clinical examination revealed bilateral internuclear ophthalmoplegia. He was treated with thiamin with a tentative diagnosis of Wernicke’s encephalopathy. MRI of brain showed hyperintense signal on T2 and FLAIR images in the peri-aqueductal grey matter which was highly suggestive of Wernicke’s encephalopathy. Later on patient revealed that the only substance he misuses in the recent past had been butane aerosol and he has not misused alcohol for many months. Conclusion: To our best knowledge this is the first case report of butane intoxication with MR findings similar to that of Wernicke’s encephalopathy. In multiple substance abusers with periaqueductal hyperintensity, the possibility of butane intoxication should be ruled out.