NOVEL THERAPY OSILODROSTAT AS AN OPTION FOR RECURRENT CUSHING’S DISEASE

Introduction: Cushing’s Disease (CD) denotes a pathologic endogenous hypercortisolism secondary to excessive adrenocorticotropic hormone (ACTH) production. Cushing Disease is a rare condition with an estimated incidence of approximately 1.2-1.4 new cases / 1,000,000 per year.
Although surgical intervention is the gold standard in the management of CD, many patients may present persistence or recurrence of disease despite surgical intervention, resulting in significant deterioration of their quality of life (QoL). For those in whom pituitary surgery is not an option or has not been curative, medical therapies have been developed. One of these newer treatments is the drug Osilodrostat, an inhibitor of the enzyme 11-beta-hydroxylase, responsible for the final step of cortisol biosynthesis in the adrenal glands. **Case Discussion:** Case of a 21 year-old female patient with history of Cushing’s Disease status post-transsphenoidal surgery (TSS) in 2017, obesity and oligomenorrhea who presented to our clinics showing signs and symptoms of hypercortisolism despite previously documented post-surgical remission in 2017. Patient reported recent development of acne, weight gain, round face, elevated blood pressure and mood changes. At the moment of evaluation, her most recent MRI showed post-surgical changes without a discrete lesion. Work-up was compatible with recurrence of Cushing’s with an ACTH at 75pg/mL (nl, 10-60pg/mL), urinary free cortisol (UFC) at 1,122mcg/24h (nl, 4-50mcg/24h) and serum morning cortisol at 24.0mcg/dL (nl, 5-20mcg/dL). In view of a negative MRI for a pituitary lesion, a Bilateral Inferior Petrosal Sinus Sampling (BIPSS) was performed. The BIPSS procedure revealed a pituitary source with a right pituitary lateralization suggestive of a right ACTH-producing tumor. The patient was consulted to Neurosurgery Service, but while waiting for repeat surgery, control of hypercortisolism was needed. The patient was started on Osilodrostat 2mg twice a day and was able to achieve normalization of UFC to 4mcg/24h, serum morning cortisol 5mcg/dL and late-night salivary cortisol (LNSC) of 30ng/dl (<90ng/dL) with significant improvement in her QoL, depression and clinical manifestations. The patient tolerated therapy well. **Conclusion:** Cushing’s Disease is a complex condition with potential serious complications if untreated. Even though TSS is the first line therapy, approximately 25% of patients show persistence of disease, and a similar proportion may experience recurrence. When surgery fails, medical treatment such as Osilodrostat, can temporarily suppress excessive cortisol production and ameliorate its clinical manifestations improving patients’ QoL while more definitive therapy is established. **Presentation:** No date and time listed