RESULTS
Twenty eight percent of patients were Asian (39/137) and enrolled in Korea (n = 14), Japan (n = 9), India (n = 7), Thailand (n = 5) and China (n = 4). Most non-Asian patients were Caucasian (n = 89/98;91%). Median (range) osilodrostat exposure from baseline to study end was 115 (1–194) weeks in Asian patients and 141 (4–245) weeks in non-Asian patients, median (range) osilodrostat dose was 3.7 (1–18) and 10.1 (1–47) mg/day, respectively. The mUFC was ≤ULN in 62% (24/39) of Asian and 68% (67/98) of non-Asian patients at W48, increasing to 68% (21/31) and 87% (65/75), respectively, at W72. Improvements in cardiovascular parameters were observed in both groups during osilodrostat treatment. Hypocortisolism-related adverse events, mostly of mild or moderate severity, occurred in 64% (25/39; n = 2 discontinued) of Asian and 50% (49/98; n = 3 discontinued) of non-Asian patients.

CONCLUSION
Beneficial effects of osilodrostat were similar in Asian and non-Asian patients in terms of biochemical control and clinical improvement, although Asian patients generally received lower doses. Osilodrostat was well tolerated in both groups.

PP-PN-06

SUCCESSFUL USE OF RITUXIMAB TO TREAT GLUCOCORTICOID RESISTANT LYMPHOPLASMACYTIC HYPOPHYSITIS: A CASE REPORT

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BACKGROUND
Lymphoplasmacytic hypophysitis is very rare and due to that its treatment is unclear. We present a case of glucocorticoid resistant lymphoplasmacytic hypophysitis successfully treated with rituximab.

CASE
A 34-year-old Sri Lankan male, presented with subacute onset severe headache and right sided visual impairment over 2 weeks without fever. He experienced nocturia with polydipsia since late adolescence which worsened with the onset of current symptoms. Later, he developed right sided parotitis. His height was 153 cm, BMI was 22.4 kg/m^2. There was complete vision loss in right visual field and bilateral optic nerve atrophy with more severe involvement on the right. He lacked secondary sexual characteristics. Hormonal evaluation demonstrated secondary hypothyroidism, hypoadrenalism, hypogonadism, low IGF-1, and normal prolactin levels. Osmolality studies confirmed the presence of diabetes insipidus. Brain MRI showed hypoplastic pituitary with contrast enhancing stalk, infiltration in the sellar, suprasellar and parasellar regions with optic nerve and cavernous sinus invasion. Parotid biopsy was normal. Inflammatory markers were elevated. Complete blood count was normal except for mild anemia. Tuberculosis, HIV, Aspergillosis, vasculitis, sarcoidosis and Langerhans Cell Histiocytosis were excluded.

Lymphocytic hypophysitis was presumed and was started on methyl prednisolone pulses with pituitary hormone replacement. Due to poor response and progression of disease, he underwent transsphenoidal biopsy which showed lymphoplasmacytic infiltration with sclerosis suggesting possible IgG4 disease. IgG4 staining was not available. Serum IgG4 levels were normal. Intravenous rituximab pulses (1 g) were given 2 weeks apart

There was marked response to rituximab. His headache settled, nasal desmopressin was tailed off and resulted in slight improvement of the vision of blind eye. Follow-up MRI scans after 5 months revealed no progression from baseline.

CONCLUSION
Rituximab may give promising results in the presence of glucocorticoid resistant lymphoplasmacytic hypophysitis.

PP-PN-07

DIAXOZIDE INDUCED THROMBOCYTOPENIA IN TREATMENT OF INSULINOMA

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BACKGROUND
Insulinoma is a rare neuroendocrine tumor. Hypoglycaemia due to insulinoma is be managed by diaxozide, a benzothiadiazine derivate with antihypertensive and hyperglycemic activities prior to definitive surgery.

CASE
A 65-year-old female was admitted with hunger pangs, sweating and giddiness after a large carbohydrate meal and weight gain of 14 kg in 2 years. On fasting, she developed symptomatic hypoglycaemia within 2 hours with random blood glucose 2.1 mmol/L, serum insulin 1213 pmol/L (N 17.8-173), C-peptide 7784 pmol/L (N 367-1469) and cortisol 598.5 nmol/L. CT pancreases showed a 5 x 5.1 x 5.3 cm irregular walled lesion with coarse calcifications at the tail of pancreas (HU 13-15) with no septation- likely insulinoma.

She was started on diaxozide for endogenous hyperinsulinemic hypoglycaemia with dose titrated up to 400 mg/day. One week after diaxozide initiation, she developed symptoms of diazoxide overdose, fever and thrombocytopenia (nadir level 10 x 10^9/L) with no source of infection. Diaxozide was stopped and she was switched to subcutaneous octreotide 100 mg TDS. Symptoms of overdosage, fever, and thrombocytopenia resolved after 4 days of stopping diaxozide. She then underwent distal pancreatectomy which was curative.

Thrombocytopenia is a rare complication of diaxozide and occurs due to platelet destruction from antibody formation. It is dose dependent and occurs 13-23 days after initiation of diaxozide and resolution will occur after discontinuing diaxozide.

CONCLUSIONS

Although thrombocytopenia is a rare complication of diaxozide, close monitoring of platelet count is needed to prevent complications.

PP-PN-08

DELAY IN GROWTH HORMONE THERAPY IS NOT DETRIMENTAL

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BACKGROUND

Pituitary hypothyroidism with growth hormone deficiency is often missed in primary care due to scarce health resources in developing countries. Well-meaning primary care with thyroxine for misdiagnosed primary hypothyroidism may be detrimental. However, the initial use of thyroxine in neonatal central hypothyroidism and growth hormone deficiency prevented long term intellectual decline even when it was inappropriately diagnosed as primary hypothyroidism.

CASE

Twin male siblings born to second degree consanguineous parents were managed by a pediatric specialist as primary hypothyroidism, requiring unusually high thyroxine doses. T4 was used to guide therapy. After consultation with an endocrinologist, the associated growth hormone deficiency was diagnosed. Pituitary structures on magnetic resonance imaging were normal.

The dose of thyroxine was beyond age-matched norms, with free T4 and free T3 values significantly high at 4 years of age. Confirmation of growth hormone deficiency was guided by hormone assay and auxology. Assistance from a growth hormone manufacturing company and government support helped manage both siblings from age 4 up to 18 years, notwithstanding limitations in periodic monitoring. The siblings are now in graduate technical college with respectable height gain.

CONCLUSION

Appropriate and timely diagnosis of pituitary hypothyroidism and associated growth hormone replacement, even in poor socioeconomic situations, can help nurture productive citizens.

PP-PN-09

AUTOIMMUNE POLYGLANDULAR SYNDROME (APS) TYPE 2 WITH CENTRAL DIABETES INSIPIDUS

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BACKGROUND

APS involves functional abnormalities in several endocrine and non-endocrine glands. Deficits may manifest over time. Screening for other manifestations is important. The major components of the syndrome are adrenal insufficiency, thyroid autoimmunity and type 1 diabetes. It is very rarely described with central diabetes insipidus (DI).

We present a very uncommon presentation of APS-2 with the onset of Addison's at age 7, developing other polyglandular associations over time and central DI in adulthood.

CASE

A 31-year-old woman with a history of Addison's disease diagnosed and treated with dexamethasone at age 7 presented with acute polyuria and polydipsia. She had negative water deprivation test according to the North Bristol protocol. Due to persistent symptoms, she was empirically commenced on desmopressin (Minirin) with good response. Testing finally diagnosed central DI: magnetic resonance imaging (MRI) demonstrated absence of the posterior pituitary bright spot and a 3 mm stalk thickening thought to be pathognomonic and possibly related to arginine vasopressin antibody status. Antibodies to thyroglobulin, thyroid receptor, ZnT8, GAD and IA2 were negative. TSH and HbA1c were normal.