Hybrid diabetes with good response to metformin in an Adolescent with polyglandular poly-endocrinopathy

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INTRODUCTION

Development of type 1 diabetes is well known in cases of type 2 Autoimmune polyglandular syndrome (APS). We describe a case of APS who developed a hybrid form of diabetes that responded to metformin therapy.

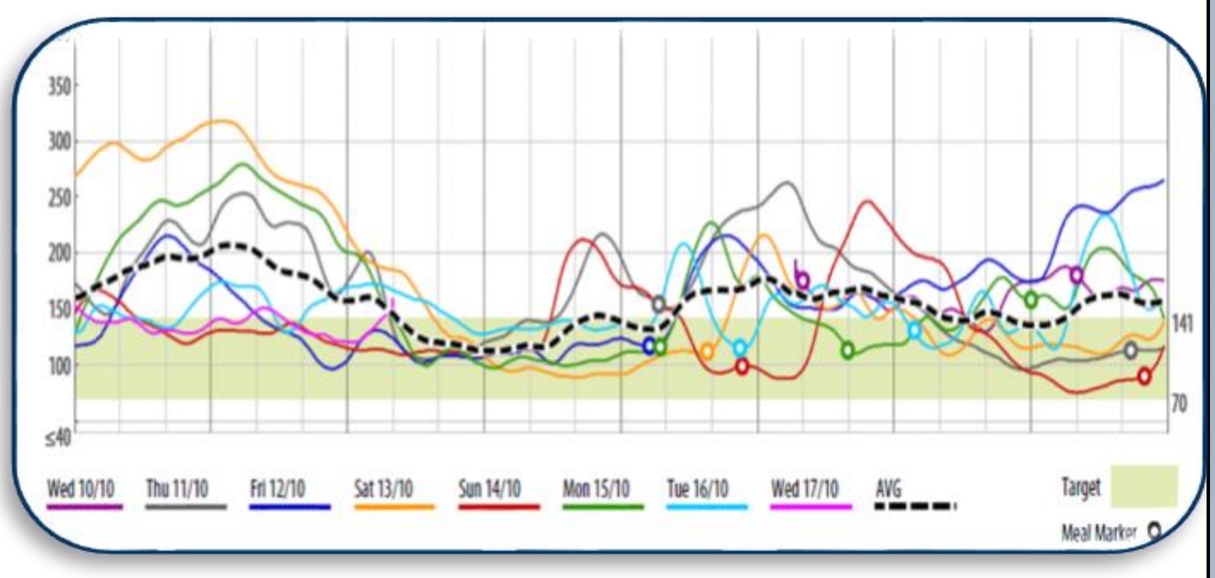
Case Study

This 18 year old male patient has been diagnosed with polyglandular endocrinopathy type 2 at the age of 13 years with primary adrenal insufficiency and autoimmune hypothyroidism . He has been on treatment with normal linear growth and pubertal development.

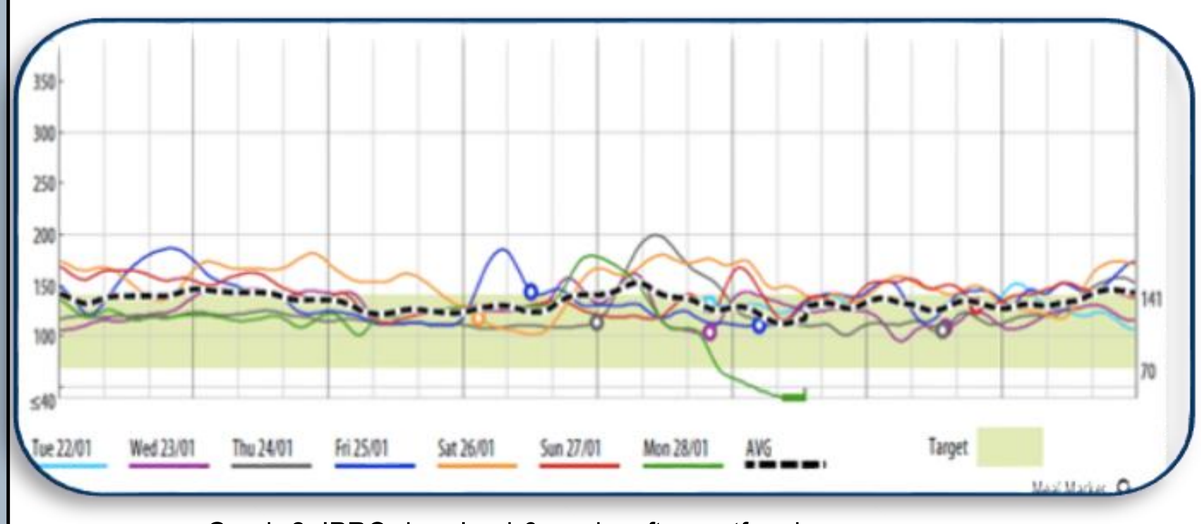
At the age of 17 years, he had excessive weight gain (BMI of 28.5 kg/m^2) and acanthosis nigricans. He had no history of polyuria or polydipsia.investigation showed : HbA1c = 7.2%, the 2 hour OGTT =14.6mmol/l , and autoantibodies screen was positive for anti GAD65 , anti-islet 2 ab and ant ZnT8 ab .

we elected to start metformin 1gm BID and observe the glycemic control as the patient showed clinical symptoms of insulin resistance .

A continuous glucose monitoring (CGM) was done with iPro for the patients in 3 stages of his follow ups showing the following charts : before treatment, at 6 weeks and , after 1 year . The following data will show the following :

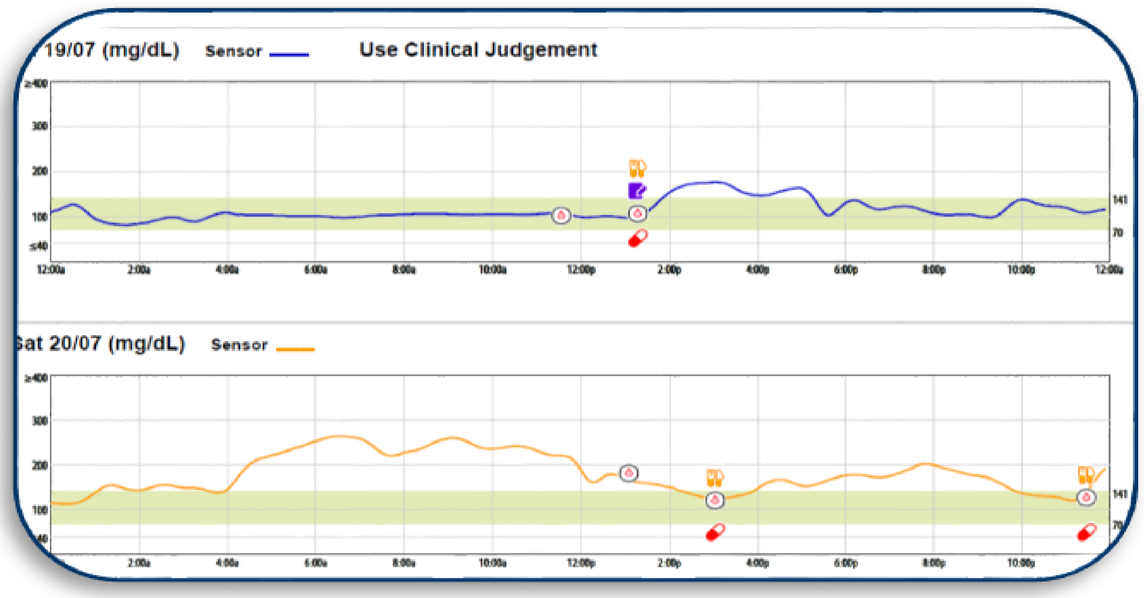


Graph 1:Hyperglycemia after the main meals as shown by IPRO CGM for one week , before starting metformin .



Graph 2: IPRO download 6 weeks after metformin use

Management



Graph 3: IPRO Daily summary :after one year of treatment , an example of the effect on the glycemic control when full dose of metformin used vs missing one dose in the in the following 24 hours .A full dose received on Thursday , with missed dinner dose on Friday .

Marker/time	0 time	1 month	1 year
Insulin pmol/l	40	NA	48
c-peptide ng/ml	2	1.77	1.77
BMI	28.5	25.5	27.2
HOMA IR	1.8	NA	2.1
HbA1c	7.2%	6.2%	6.9%

Table 1:the relationship with patient`s HbA1c , insulin reserve and resistance with BMI in our patient

Discussion and conclusion

Patients with APS-2 are characterized by at least two of the following three endocrinopathies: type 1 diabetes, autoimmune thyroid disease, and Addison's disease. The DR3-DQ2/DRB1*04:04-DQ8 genotype has been associated with type 1 diabetes in patients with APS 2.

This is the first report that describes the occurrence of hybrid diabetes in a case of APS 2 , in which the patient responded well after one year of metformin . The compliance on metformin and BMI proved to have the major effect in the glycemic control as in other patients with hybrid diabetes .

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