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Post-prandial hyerinsulinaemic hypoglycaemia post-esophageal surgery in children

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Background

- Post-prandial hyperinsulinaemic hypoglycaemia (PPHH) or dumping syndrome is a recognized complication of various gastric surgeries. There are very few paediatric case reports to confirm PPHH post esophageal repair.
- In children, PPHH has been described almost exclusively as a postoperative complication of gastric fundoplication [1,2].
- A number of studies using gastric scintigraphy, manometry, or electrogastrography have shown that abnormal gastric emptying is frequent in patients treated surgically for Eosophageal atresia (EA) [3,4]

Case 1

- 6 month old female baby born at 38^{+3} weeks gestation, by elective C-section, with birth weight of 2040 grams was diagnosed with Wolf-Hirschhorn syndrome.
- Diagnosed antenatally with tracheo-oesophageal fistula, that was surgically repaired on day 2 of life and subsequently requiring monthly oesophageal dilatations.
- Developed episodes of hypoglycaemia post-operatively, following oesophageal balloon dilation.
- Further investigations have been consistent with dumping syndrome
- Initially responding to Diazoxide (7mg/kg/day) and continuous feeds.
- Developed Pulmonary Hypertension and subsequently Diazoxide stopped
- She is currently stable on continuous feeds.

Case 2

- 2 year old boy, born at 38 weeks gestation, with an antenatal diagnosis of long segment oesophageal atresia (without fistula).
- On day 3 of life, a fashioned gastrostomy was performed to facilitate feeding. During the first year of life, he required six oesophageal dilatations for the strictures and at that point he had normal blood glucose levels.
- At the age of 11 months he presented with a hypoglycaemic seizure
- The biochemical investigations confirmed the diagnosis of Dumping syndrome
- Initially, he was tried on Acarbose and Diazoxide, but proven to be unresponsive. Finally, he was managed with continuous feeds over seventeen hours and fasting up to eight hours.
- He is currently stable has been stable on continuous feeds

Results

CLINICAL DATA AT REFERRAL	Case 1	Case 2
Age of presentation	6 months	11 months
Symptoms at presentation	Episodes of hypoglycaemia	Hypoglycaemic seizure
Type of GI malformation	Tracheo-oesophageal fistula, that was	Long segment oesophageal atresia (without fistula).
Age at surgery	Day 2 of life	Day 3 of life
Type of surgery	Surgically repaired Monthly oesophageal dilatations	Fashioned gastrostomy Required six oesophageal dilatations for the strictures the 1st year of life
Dysmorphic features	Wolf-Hirschhorn syndrome	None
Response to Diazoxide (maximal dose)	Responsive (7 mg/kg/day) Discontinued (Pulmonary hypertension)	Unresponsive (5 mg/kg/day) Discontinued
Response to Octreotide (maximal dose)	Not required	Not Required
Response to other treatment (maximal dose)	No	Unresponsive to Acrabose (20gr TDS)
Response to Continuous Feeds	Yes	Yes
Current Treatment	▪ Overnight: 20ml/hour of continuous feed for 20 hours ▪ Daytime: Two breaks of two hours while she takes solid food on demand.	▪ Overnight: 65ml/hour of continuous feed for 12 hours ▪ Daytime: Two bolus feeds of 65ml given over 1 hour. Solids food and orange juice on demand

What is already known on this topic

- PPHH manifests as nonspecific symptoms after eating, including refusal to eat, postprandial nausea, retching, pallor, lethargy, diaphoresis, watery diarrhea
- In childhood, the surgical treatment of gastroesophageal reflux is by far the main cause of PPHH
- Other causes of PPHH in children are very rare and include congenital microgastria, partial or total gastrectomy, accidental intraduodenal or jejunal administration of bolus feeding, or inadequate meals with high osmolarity, as well as rare cases of generalized autonomic dysfunction .
- In children with esophageal atresia (EA), all reported cases of PPHH have so far been related to the surgical treatment of associated gastroesophageal reflux

Conclusions

- ✓ PPHH syndrome following oesophageal surgeries for gastro oesophageal reflux and oesophageal atresia are quite common
- ✓ PPHH syndrome remains an unrecognized complication following oesophageal atresia in paediatric population

What this study adds

- ✓ PPHH is not well known in children undergoing oesophageal atresia repair and hence it is important to be aware of this rare complication and screen these patients when are symptomatic regularly.
- ✓ Continuous feeds might be the only option for PPHH if unresponsive to medical therapy
- ✓ PPHH can get milder over time, as proven by the second case

References

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