Post-prandial hyperinsulinaemic hypoglycaemia after oesophageal surgery in children

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Introduction

- Post-prandial hyperinsulinaemic hypoglycaemia (PPHH) or late dumping syndrome is a recognised complication of various gastric surgeries.
- In children, PPHH has been described almost exclusively as a postoperative complication of gastric fundoplication.
- There are very few paediatric case reports to confirm PPHH post esophageal repair, without any known precipitating factors such as gastroesophageal reflux surgery or associated microgastria.

Case 1:

- 2-year-old boy diagnosed with oesophageal atresia at birth
- Surgically repaired requiring six oesophageal dilatations the first year of life.
- At 11-months of age had hypoglycaemic seizures and PHHH confirmed
- Acarbose and diazoxide trials failed.
- Managed with 17-hours continuous gastrostomy feeds.
- Currently, he is 28-months-old with euglycaemia on daytime bolus gastrostomy feeds and overnight 12-hours continuous gastrostomy feeds.

Case 2

- 6-month-old girl diagnosed with Wolf-Hirschhorn syndrome and tracheo-oesophageal fistula
- Surgically repaired, requiring monthly oesophageal dilatations.
- At 5-months of age reported to have hypoglycaemia and PPHH confirmed.
- Responded to diazoxide and continuous/bolus nasogastric tube feeds, **but developed pulmonary hypertension** possibly diazoxide-induced.
- Normoglycaemia was secured via 20-hours continuous gastrostomy feeds.

Clinical features, biochemical investigations and treatment response in two patients with PPHH

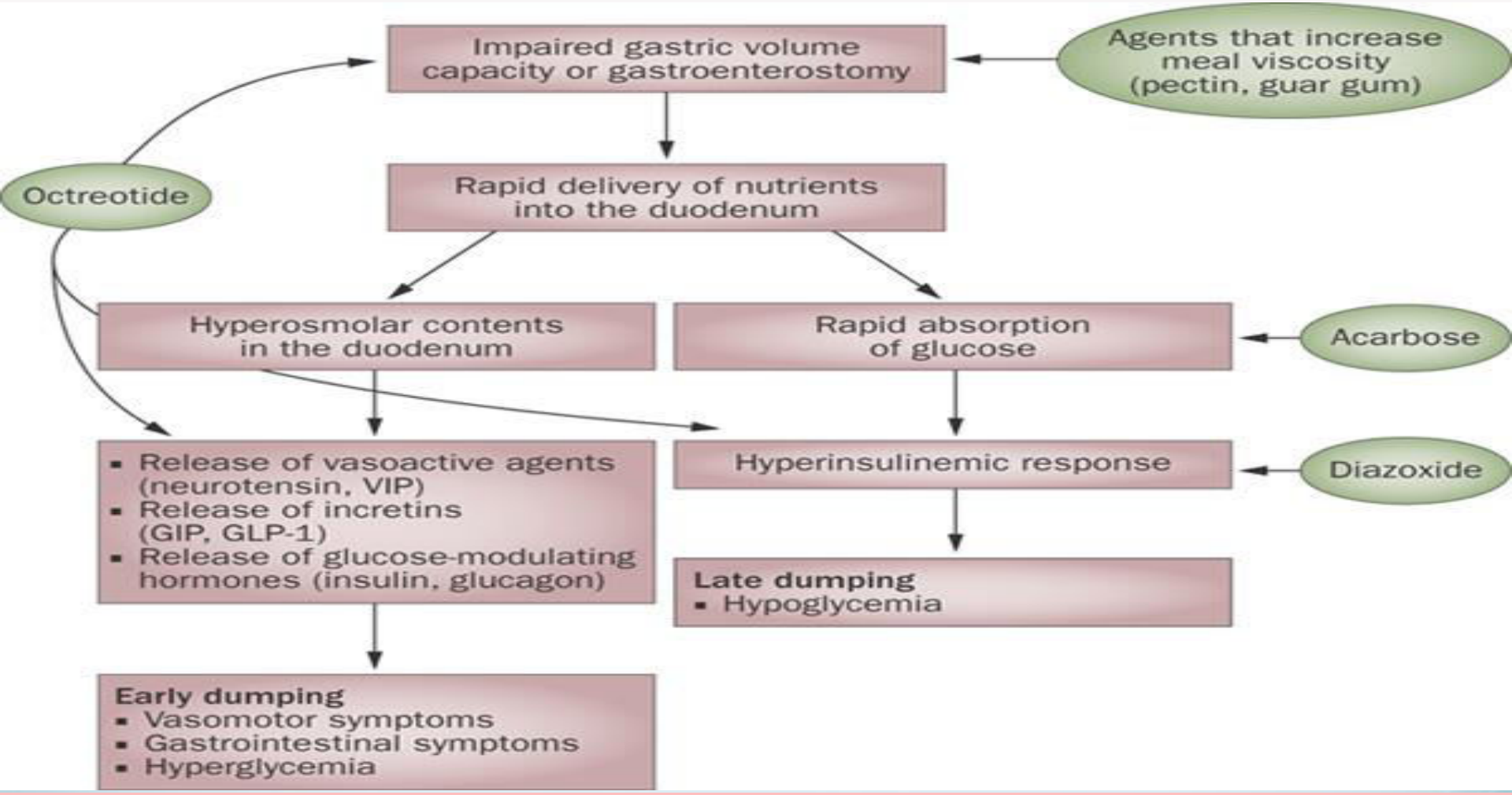
	Case 1	Case 2
Type of gastrointestinal malformation	Oesophageal atresia (Long gap) No fistula	Oesophageal atresia Tracheoesophageal fistula
Age at surgery (days)	90	2
Type of surgical procedures	Delayed primary repair following Replogle suction/irrigation of the upper pouch, gap assessments, gastrostomy insertion. Six oesophageal dilatations the first year of life	Closure of tracheo-esophageal fistula Repair of oesophageal atresia Monthly oesophageal dilatations
Dysmorphic features	None	Wolf-Hirschhorn Syndrome
Age at presentation	11 months	5 months
Symptoms at presentation	Hypoglycaemic seizure	Incidental episode of hypoglycaemia
Feeding plan at presentation	3 hourly bolus feeds of 100ml (CHO content: 7.7%)	1-2 hourly bolus NGT feeds (CHO content: 7.7%)
Provocation test. Time of hypoglycaemia. Results.	Mixed meal test 90 minutes capillary glucose 2.6mmol/L Insulin 3.0mU/L	Mixed meal test 60 minutes capillary glucose 1.9mmol/L insulin 17.9mU/L
Response to Acarbose (max dose)	Unresponsive (25grams every 8 hours)	Not tried (due to maternal concerns regarding side effecrs)
Response to Diazoxide (max dose)	Unresponsive (5 mg/kg/day)	Responsive (7 mg/kg/day) Discontinued-Pulmonary hypertension
Response to Continuous Feeds	Yes	Yes
Feeding regimen	18 hours continuous feed 3hourly breaks twice a day	19 hours continuous feeds Very small oral intake day time 2.5hourly breaks twice a day

PPHH

- PPHH or late dumping syndrome: Non-specific symptoms 1-3 hours after a meal, with signs and symptoms of hypoglycaemia.

- Causes in childhood:
  - Surgical treatment of gastroesophageal reflux
  - Abnormal gastrointestinal anatomy: congenital microgastria, partial or total gastrectomy, accidental intraduodenal or jejunal administration of bolus feeding, or inadequate meals with high osmolality, generalized autonomic dysfunction.
  - Idiopathic PPHH in children without a known underlying cause
  - Post-oesophageal repair without any known precipitating factor

Pathophysiology of dumping syndrome
Mode of action of different therapeutic agents



Tack, J. *et al.* (2009) *Nat. Rev. Gastroenterol. Hepatol.* doi:10.1038/nrgastro.2009.148

- ✧ Change of feeding regimens (composition, volume, rate)
- ✧ May require continuous feeds

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Conclusion

- ✓ PPHH may be an under-diagnosed complication in children undergoing surgery for oesophageal atresia.
- ✓ These children must be monitored closely for symptoms of hypoglycaemia and if there are concerns must be screened for possible PPHH.
- ✓ Our cases demonstrate that continuous feeding regimens might be the only therapeutic option, until PPHH gradually lessens in intensity over time.