Anesthetic management of a case of nesidioblastosis

Sir.

Nesidioblastosis is a rare disorder of autonomous insulin secretion from the exocrine glands.^[1] It usually presents during early neonatal or infantile period with manifestation of signs and symptoms associated with hypoglycemia. We present here the anesthetic management in a difficult case of nesidioblastosis.

A 6-month-old 8 kg male baby was diagnosed to have hypoglycemia (random blood sugar [RBS] <25 mg%) 2-h postbirth and was treated with intravenous (IV) glucose. He was born to a second gravida mother by cesarean section due to cephalopelvic disproportion at 34-week gestation. In the present admission, he had RBS <20 mg% and MRI abdomen showed hypertrophy of pancreas. A diagnosis of nesidioblastosis was made, and an elective near total pancreatectomy was planned. He was put on syrup diazoxide and octreotide. He was started 12.5% dextrose IV at the rate of 32 ml/h and continued till he was shifted to operating room. On the day of surgery, premedication in the form of syrup midazolam (0.5 mg/kg) was administered orally. His preinduction RBS was 83 mg/dl. Anesthesia was induced with IV fentanyl 20 µg and thiopentone 30 mg and was maintained on isoflurane (1.5%–2.0%) in N₂O:O₂ (50:50). Intraoperative analgesia and muscle relaxation were achieved with boluses of fentanyl (0.5 mcg/kg) and atracurium (0.5 mg/kg). IV fluids were changed to 5% dextrose in 0.3% normal saline. After induction, his RBS was 214 mg/dl, which rose to level of 317 mg/dl. Hence, a single injection of 3 units of plain insulin was administered IV, also fluid was changed from 5% dextrose to Ringer's lactate, and after this, RBS decreased to 150 mg/dl within 30 min. Perioperative analgesia was managed with combination of morphine and bupivacaine caudally and IV fentanyl. Postoperatively, RBS fluctuated between 150 and 210 mg/dl and was managed with insulin as required. He was discharged after 10 days when the RBS readings became stable without insulin.

Hypoglycemia in infancy is defined as a blood sugar level of <30 mg% in the first 3 days and 40 mg% thereafter in the term infant, and nesidioblastosis is the most common cause of persistent hyperinsulinemic hypoglycemia.^[1,2] Genetic mutation with dependent potassium channel of ATP is considered responsible for it.^[3] Usually, it presents

with convulsion just after birth secondary to severe hypoglycemia.[4] Subtotal to near total pancreatectomy is indicated as a matter of urgency to decrease the amount of circulating insulin. [4] Various measures to tackle hypoglycemia have been advocated which include concentrated glucose hydrocortisone 5 mg/kg 12 hourly IV, glucagon 0.1 mg/kg, and diazoxide 10-25 mg/kg/day.[4] Some very important precautions that need to be taken include premedication by administering oral midazolam due to its anticonvulsive effect and avoiding inhalational agent such as sevoflurane which may provoke convulsions.[5,6] We had avoided sevoflurane and used IV agents such as thiopentone and fentanyl instead as thiopentone has a better anticonvulsant action. This important anesthetic consideration has not been discussed previously in the literature in the management of nesidioblastosis. Hence, we would like to propone that frequent perioperative RBS monitoring, timely treatment of hypo- or hyperglycemic episodes, adequate fluid management, and use of appropriate anesthetic drugs are pivotal in the management of nesidioblastosis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's parents have given their consent for their child's images and other clinical information to be reported in the journal. The patient's parents understand that their child's name and initial will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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References

- Schiller M, Krausz M, Meyer S, Lijovetzky G, Landau H. Neonatal hyperinsulinism – Surgical and pathologic considerations. J Pediatr Surg 1980;15:16-20.
- Bellwoar C, Schwartz R, Stayer S. Anaesthetic management of a neonate with nesidioblastosis. Paediatr Anaesth 1996;6:61-3.
- Mali M, Bagry H, Vas L. Anaesthetic management of a case of nesidioblastosis for subtotal pancreatectomy. Paediatr Anaesth 2002;12:80-4.
- García-Santos EP, Manzanares-Campillo Mdel C, Padilla-Valverde D, Villarejo-Campos P, Gil-Rendo A, Muñoz-Atienza V, et al. Nesidioblastosis. A case of hyperplasia of the islets of langerhans in the adult. Pancreatology 2013;13:544-8.
- Aynsley-Green A, Polak JM, Bloom SR, Gough MH, Keeling J, Ashcroft SJ, et al. Nesidioblastosis of the pancreas: Definition of the syndrome and the management of the severe neonatal hyperinsulinaemic hypoglycaemia. Arch Dis Child 1981;56:496-508.
- Nieminen K, Westerèn-Punnonen S, Kokki H, Yppärilä H, Hyvärinen A, Partanen J, et al. Sevoflurane anaesthesia in children after induction of anaesthesia with midazolam and thiopental does not cause epileptiform EEG. Br J Anaesth 2002;89:853-6.

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