Hyperbaric oxygen treatment in bilateral orchiopexy and post-circumcision haematoma in a thrombocytopenic patient with Noonan syndrome

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Abstract

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Hyperbaric oxygen treatment (HBOT) can be utilised for necrotising soft tissue infections, clostridial myonecrosis (gas gangrene), crush injuries, acute traumatic ischaemia, delayed wound healing, and compromised skin grafts. Our case was a 17-month-old male patient with Noonan syndrome, idiopathic thrombocytopenic purpura, and bilateral undescended testicles. Haematoma and oedema developed in the scrotum and penis the day after bilateral orchiopexy and circumcision. Ischaemic appearances were observed on the penile and scrotal skin on the second postoperative day. Enoxaparin sodium and fresh frozen plasma were started on the recommendation of haematology. Hyperbaric oxygen treatment was initiated considering the possibility of tissue necrosis. We observed rapid healing within five days. We present this case to emphasise that HBOT may be considered as an additional treatment option in patients with similar conditions. To our knowledge, no similar cases have been reported in the literature.

Introduction

Noonan syndrome is an autosomal dominant syndrome occurring in 1 in 1,000–2,500 live births, attributed to mutations in the PTPN11 gene. It is characterised by short stature, a low hairline on the nape of the neck, a webbed neck, cubitus valgus, pectus excavatum, and cardiac anomalies. Additionally, haematological issues have been documented in affected individuals, with bleeding disorders being of particular concern. Thrombocytopenia, Von Willebrand disease, and platelet dysfunction may manifest, leading to varying degrees of bleeding abnormalities. Such haemostatic system disorders can provoke post-operative complications including haematoma formation.

Case report

Informed consent was obtained from the patient's parents for the publication of the case report and photographs of the patient.

A 17-month-old male patient with Noonan syndrome was admitted to the paediatric urology clinic for bilateral undescended testicles and circumcision. Physical examination revealed characteristic features of Noonan syndrome, including low-set ears, a high palate, almond-shaped eyes, and a pointed chin. On urological examination, both testicles were palpable at the entrance of the inguinal canal. Scrotal ultrasound showed that the right testicle was 11 x 5 x 7 mm (0.26 ml), and the left testicle was 9 x 5 x 11 mm (0.25 ml), with no significant difference in parenchymal echogenicity between them. The patient was under haematological follow up due to idiopathic thrombocytopenic purpura (ITP). Chromosomal analysis confirmed a 46XY karyotype with a mutation in the PTPN11 gene.

Bilateral orchiopexy and circumcision were performed uneventfully. On the first postoperative day, the patient experienced a minor haemorrhage at the circumcision site, which was managed with local application of tranexamic acid. Although the bleeding ceased upon follow-up, haematoma and oedema developed in the scrotum and penile skin. Dressings were applied using soft paraffin-saturated gauze with chlorhexidine acetate BP. Ischaemic changes were noted on the penile skin and scrotum on the second postoperative day (see Figures 1 and 2).

Hyperbaric oxygen treatment (HBOT) was initiated promptly. The patient received 100% oxygen at 243 kPa (2.4 atmospheres absolute) for 120 minutes per session.

Figure 1
Postoperative day one; haematoma initially appeared

Figure 2
Postoperative day two; progression to suspicious necrosis

Figure 3
Postoperative day three; first day of hyperbaric oxygen treatment

Figure 4
Postoperative day four and second day of hyperbaric oxygen treatment; a significant improvement was observed in the haematoma

After the initial HBOT session, three additional sessions were completed within the first 24 hours. Subsequent improvement in ischaemic changes was observed after the fourth session. Hyperbaric oxygen was continued once daily for five days thereafter (see Figures 3–7). Concurrently, the patient was given 10 ml·kg^{-1} fresh frozen plasma and 1 mg·kg^{-1}·day^{-1} enoxaparin sodium (low molecular weight heparin) for five days. The patient was discharged at the seventh day with complete wound healing (Figure 8)

Discussion

Hyperbaric oxygen treatment involves administration of 100% oxygen in a closed chamber at pressures higher than sea level, delivered via a mask, hood, or endotracheal tube.[1] It increases the dissolved oxygen content in the blood plasma and provides hyperoxygenation to tissues that have increased oxygen demand or reduced supply. This increase in blood oxygen content and partial pressure also compensates for arterial vasoconstriction caused by hyperoxia. It has been shown to promote vascular proliferation by increasing vascular endothelial growth factor elaboration and stem cell mobilisation as well as enhancing host defense against infections and regulating the anti-inflammatory response.[2,3]

In paediatric patients, HBOT is used for necrotising soft tissue infection, clostridial myonecrosis (gas gangrene), crush injuries, and acute traumatic ischaemia. One study[4] reported that HBOT was associated with favorable functional and cosmetic outcomes in penile repair after urethral and glans penis reconstruction following penile amputation, while another[5] demonstrated positive results with HBOT following microsurgery in a replanted penis after penile amputation.

Figure 5
Postoperative day five and third day of hyperbaric oxygen treatment

Figure 6
Postoperative day six and fourth day of hyperbaric oxygen treatment

Figure 7
Postoperative day seven and fifth day of hyperbaric oxygen treatment

Figure 8
Postoperative day eight; complete resolution of haematoma was achieved

In the present case, HBOT was initiated to overcome the ischaemic complication of scrotum oedema, ecchymosis and haematoma of the penis and scrotum. We achieved complete tissue recovery without necrosis. By presenting this case, we aim to contribute to the existing literature on the use of HBOT in similar clinical scenarios. We believe that the anti-oedema effects of HBOT improved oxygen levels allowing complete recovery without tissue loss.

Conclusions

We cannot be certain that HBOT materially altered the outcome in the present case. Nevertheless, the contemporaneous improvement seen after institution of HBOT suggests there may have been some benefit. Moreover, HBOT might be considered in treatment of similar cases which, due to their sporadic occurrence, are never likely to be the subject of large studies.

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