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Anaesthesia for Abscess Drainage in a Six-Month-Old Male with CLOVES Syndrome

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Abstract

The term CLOVES stands for congenital lipomatous overgrowth, vascular malformations, epidermal nevi, and spinal/skeletal anomalies. These are the clinical features associated with the syndrome. CLOVES syndrome is a very rare disorder with an estimated incidence rate of less than 1:1,000,000. Its pathogenesis involves postzygotic somatic gain-of-function mutations of the PIK3CA gene on chromosome 3q26.32. It is characterized by a congenital (early) childhood onset. In this report is a case study of a six-month-old baby with features of CLOVES who presented to the hospital with multiple abscesses on the right arm and shoulder. Incision and drainage were done under general anaesthesia and endotracheal intubation, which was uneventful contrary to expectations of difficult intubation and ventilation, as well as coagulopathies and cardiovascular challenges. The patient was then referred for further treatment.

Keywords

CLOVES, Syndrome, Congenital, Male, Anesthesia

1. Introduction

The term CLOVES stands for congenital lipomatous overgrowth, vascular malformations, epidermal nevi, and spinal/skeletal anomalies. These are the clinical features associated with the syndrome. CLOVES syndrome was recently included in the PIK3CA-related overgrowth spectrum (PROS), a new entity that includes all the overgrowth syndromes characterized by PIK3CA-activated mutations [1] [2].

CLOVES syndrome is an extremely rare disorder, with very few cases reported so far [3]. Its pathogenesis involves postzygotic somatic gain-of-function mutations of the PIK3CA gene on chromosome 3q26.32. This provokes a disruption in

the downstream regulation of the PI3K/AKT/mTOR signal transduction pathway [4]. CLOVES syndrome is characterized by a congenital (early) childhood onset. It appears to affect males and females equally, irrespective of ethnicity, and its incidence rate is estimated to be less than 1:1,000,000 [5]. Reports of anesthetic management for any procedure in this very rare group of patients are scarce in the literature.

2. Case Presentation

A six-month-old baby with features of CLOVES presented to the hospital with multiple abscesses on the right arm and shoulder. Incision and drainage of the abscesses were performed under general anaesthesia.

The mother noted that the child was born with these disproportional features, but they had developed increasingly over time. The baby appeared to have an unusual body structure, with abnormally overgrown tissues over the chest, abdomen, and both upper and lower limbs (**Figure 1**). The baby also had abnormally large hands and feet (**Figure 2**). There were pigmented skin lesions mostly suggestive of capillary malformation with ulcerations, and the mother reported them to be haemorrhagic sometimes (**Figure 3**). The feel of the unusual overgrowth tissue would be best described as a "balloon of water".

Though the cardiovascular and respiratory examination was difficult to access due to overgrown truncal subcutaneous tissue, they appeared quite normal; the apex was not displaced on palpation, heart sounds were audible and normal with no audible murmurs. The room air oxygen saturation was 98%. No organomegaly was noted on abdominal examination. There was a hyperpigmented lesion on the left thigh with ulceration that the mother reported to have been bleeding. There were also abscesses noted on the right shoulder (**Figure 4**).

Figure 1. Lipomatous overgrowth of the chest, upper limbs, and abdomen.

Figure 2. Tissue overgrowth of the feet.

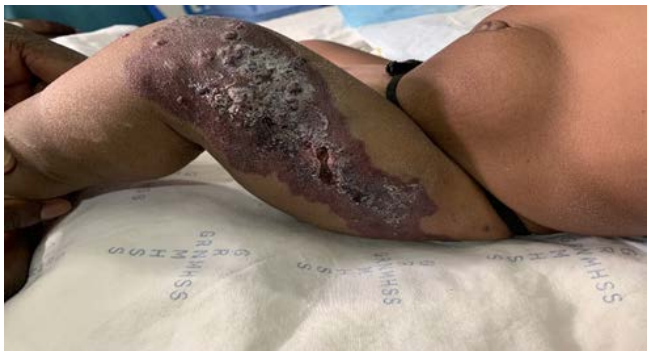

Figure 3. Hyperpigmented skin lesions with ulcerations.

Figure 4. Subcutaneous abscesses for incision and drainage.

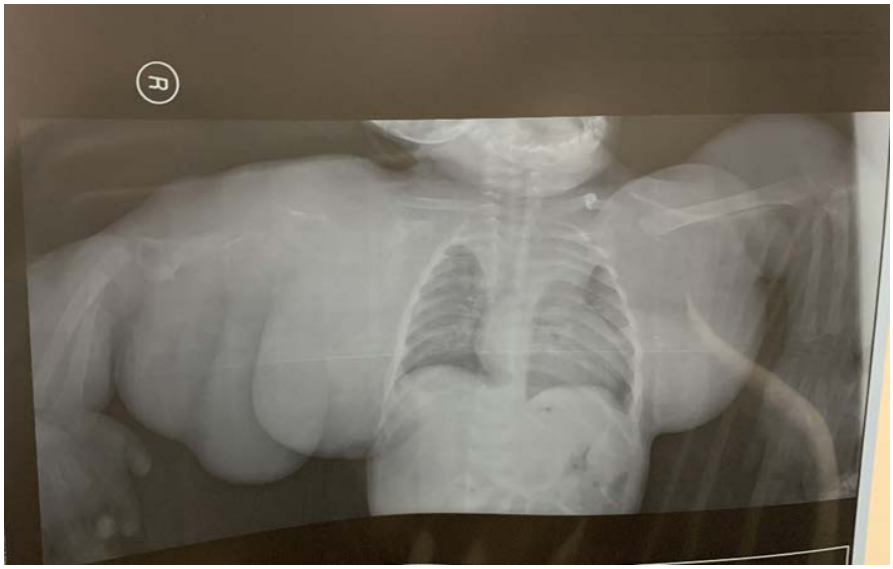

Figure 5. Chest X-ray of the patient with increased soft tissue of the chest and upper limbs.

The laboratory investigations performed included a full blood count, plasma electrolytes, and a renal function test, which yielded normal values except for a low hemoglobin level of 9.3 g/dl, attributable to active bleeding from the hyperpigmented skin lesion on the leg.

Anaesthesia was induced using sevoflurane inhalation to facilitate placement of venous access, which was difficult because of the excessive adipose tissue deposition in the limbs. Lean weight for age was used for drug calculation. This was a critical safety consideration in patients with significant lipomatous overgrowth to prevent drug overdose. Three different sizes of tracheal tube were prepared in view of the disproportionate size of the patient and anticipated difficult airway; however, an age-appropriate tracheal tube size 3.5 cuffed was placed with ease before the procedure was commenced. The inhalational anaesthetic, sevoflurane was able to sedate and obtund airway reflexes for endotracheal intubation without the need for muscle relaxant. Standard monitoring using a pulse oximeter, capnography, 3-lead electrocardiogram, non-invasive blood pressure, and temperature was done. The cuff of the blood pressure monitor was placed on the leg, away from the lipomatous tissue overgrowth in the upper limbs, for a more accurate reading. Despite the weight and size of the patient, the blood pressure was within the range for the patient's age. The ventilation mode utilised was the pressure-controlled mode in view of the reduced chest wall compliance due to the truncal tissue overgrowth as shown in Figure 5 above.

The procedure (incision and drainage of the abscess) lasted for about 40 minutes under uneventful anesthesia and monitoring, before the patient was woken up successfully and transferred to the recovery room on supplemental oxygen of about 2 L/min by nasal prong. Monitoring continued until the child was fully conscious and alert with oxygen saturation on room air that was equal to the preoperative value of 98%. Drugs used for anesthesia included glycopyrrolate,

atracurium, fentanyl, and paracetamol. He was subsequently referred to another center for further management.

3. Discussion

A definitive diagnosis of CLOVES syndrome can be established by genetic testing, which guarantees the identification of the CLOVES syndrome-specific underlying mutations. If a CLOVES syndrome diagnosis cannot be established by genetic sequencing or NGS, then the diagnosis can be established clinically [6].

Very few cases of anaesthesia for patients with CLOVES syndrome have been published. Patients have many associated features which may affect the administration of anaesthesia. Vascular malformation, abnormal subcutaneous tissue overgrowth, skeletal and muscular malformation, neural defect, and visceral anomalies are all associated with this syndrome. Patients with CLOVES syndrome majorly do not have hematopoietic, cardiovascular, and gastrointestinal anomalies, but the most common features seen include congenital lipomatous overgrowth, vascular anomalies, epidermal nevi, and skeletal deformities [2].

Generally, there is no definitive cure for CLOVES syndrome, and management is primarily geared toward improving the quality of life. Surgical debulking for lipomatous masses can be technically challenging, associated with high morbidity and high rates (~100%) of postoperative recurrence [7]. Thus, the decision for surgical intervention demands that both parties (that is, the parents and the surgeon) thoughtfully weigh the risks and benefits prior to embarking on the surgical intervention. The patient in this case had incision and drainage of lipomatous necrotic tissue in the trunk.

Depending on the organ involved, the possible complications of vascular anomalies include visceral disorders, pain, breathing difficulties, hemoptysis, and intestinal bleeding or obstruction, among others. Pharmacologic mammalian target of rapamycin (mTOR) inhibitors, such as sirolimus, have been shown to exhibit therapeutic responses with acceptable toxicity profiles in patients with complex vascular anomalies [8]. However, some grey areas regarding the pharmacokinetics of sirolimus still exist, and resolution has only been reported in a low percentage of patients in the literature [9].

The clinical complexity of CLOVES syndrome requires multidisciplinary involvement from paediatric surgeons, anaesthetists, dermatologists, orthopedic surgeons, neurologists, and radiologists. These patients undergo repeated surgical interventions that require anaesthesia.

The presence of vascular malformations, a constant feature in these patients, and excess adipose tissue made vascular access difficult in this patient. Postoperative anticoagulant therapy is essential in patients with CLOVES to reduce the risk of venous thrombosis [10]. Most of the overgrowth of tissue involves the somatic tissues, so airway involvement is rare as it does not have fatty tissue proliferation. Though the presence of other coexisting congenital vascular and lymphatic malformations can affect the neck and thoracic region, making intubation difficult, in

the index patient, endotracheal intubation was easy.

In patients with fatty tissue infiltration of the pleural region, or excessive truncal adipose tissue overgrowth, there is an accompanying restrictive thoracic cavity that would lead to breathing difficulties. Although respiratory difficulty was not observed in this patient, in the immediate postoperative period, he required oxygen supplementation for up to 30 minutes before maintaining adequate saturation in room air. This may be a result of the increased work of breathing due to reduced chest wall compliance leading to hypoxaemia. Close monitoring of oxygen saturation and a low threshold for supplemental oxygen administration are therefore recommended during the early recovery phase from anaesthesia in this category of patients.

Following the tissue overgrowth, the paediatric patients often have more than the estimated weight for their age. In this patient, the estimated weight based on age was utilized in calculating drug and fluid replacement to avoid overdose. The endotracheal tube size also corresponded with that of other children of their age, which suggested that the airway is spared from the morphological overgrowth.

4. Conclusion

The CLOVES syndrome patients often undergo repeated surgeries to correct the soft tissue overgrowth and other complications associated with the disease. While the airway may be spared, significant challenges related to vascular access, drug dosing, and potential respiratory compromise due to truncal mass effect remain important anaesthetic considerations. Oxygen supplementation is needed in situations where reduced chest wall compliance leads to hypoxaemia in the early recovery period.

Conflicts of Interest

All authors declared that they have no conflicts of interest.

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