ORIGINAL RESEARCH PAPER
Volume - 13 | Issue - 02 | February - 2024 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

A RARE PRESENTATION OF DISSEMINATED LANGERHANS CELL HISTIOCYTOSIS IN A 2-YEAR-OLD CHILD: AN ANAESTHETIC AND SURGICAL MANAGEMENT CASE REPORT.

Anaesthesiology

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ABSTRACT

In this case report, we present a patient with Langerhans cell histiocytosis (LCH) who presented with an axillary abscess. LCH, an idiopathic condition characterised by abnormal proliferation of Langerhans cells, can manifest with varied systemic involvement, affecting organs such as bone marrow, lungs, liver, spleen, lymph nodes, gastrointestinal tract, and the pituitary gland. The patient underwent a biopsy and drainage procedure for the axillary abscess, posing challenges to the anaesthetist due to the potential multi-system nature of the disease.

KEYWORDS

Langerhans cell histiocytosis, Difficult airway, Nerve block, Erector spinae, Fascia iliaca, Diabetes insipidus.

INTRODUCTION

In this case study, we describe a patient diagnosed with Langerhans cell histiocytosis (LCH) who presented with an abscess in the axillary region. LCH is an idiopathic condition characterised by abnormal proliferation of Langerhans cells, and it can manifest with diverse systemic involvement. The patient underwent a biopsy and drainage procedure for the axillary abscess, posing challenges to the anaesthetist due to the potential multi-system nature of the disease. Thorough preoperative assessment, meticulous anaesthetic planning, and close postoperative monitoring were crucial to address the complexities associated with LCH and ensure optimal patient care. This case underscores the significance of a multidisciplinary approach in the management of patients with LCH undergoing surgical interventions.

CASE REPORT

We report the case of a 2-year-old child weighing 10 kg who presented with multiple skin lesions over the trunk, back, neck, forehead, and scalp, accompanied by cervical lymphadenopathy and a large, inflamed, and tender axillary swelling diagnosed as an abscess. Further investigations revealed disseminated Langerhans cell histiocytosis (LCH) involving the scapula, skull, lungs, and liver. The child, scheduled for drainage of the axillary abscess and bone marrow biopsy for reconfirmation of the diagnosis, presented preoperatively with severe anemia, elevated white blood cell count, and increased C-reactive protein levels. A suspected difficult airway was identified due to enlarged lymph nodes around the neck. Preoperative optimisation included blood transfusion, and the patient underwent surgery under general anaesthesia with an uncuffed endotracheal tube. Landmark-guided erector spinae plane block at T4 level and fascia iliaca block were administered for analgesia, along with intravenous paracetamol. The procedure was uneventful, and the child was extubated at the end, transitioning to the post-anaesthesia care unit. The patient remained comfortable and pain-free for approximately 12-13 hours, after which intravenous analgesics were initiated. Subsequent biopsy results confirmed LCH, leading to the patient's referral for chemotherapy. This case highlights the importance of comprehensive peri-operative management in paediatric patients with disseminated LCH, incorporating a multidisciplinary approach for successful surgical outcomes and postoperative care.

DISCUSSION

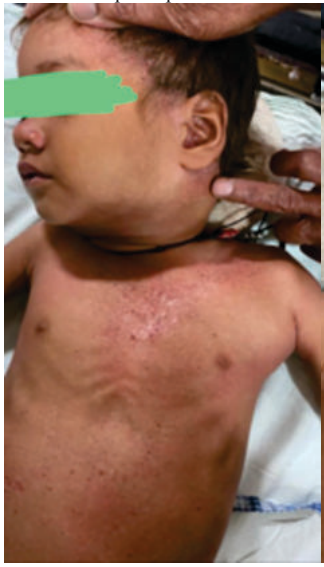
Langerhans cell histiocytosis (LCH) is an uncommon disease with potential multisystem involvement, posing challenges for both prognosis and anaesthetic management[1]. In our case, the patient presented with bony lesions, emphasising the need for meticulous care during patient transfer and positioning due to the risk of bone pain and pathological fractures. The potential for oral bleeding during intubation underscores the importance of gentle laryngoscopy in order to minimise complications.

One notable aspect of LCH is its capacity to affect the hypothalamo-pituitary-adrenal (HPA) axis[2], leading to complications such as diabetes insipidus and hypernatremia. Although these manifestations were not present in our case, diligent monitoring of blood sugar and urine output is imperative in patients with suspected or confirmed LCH.

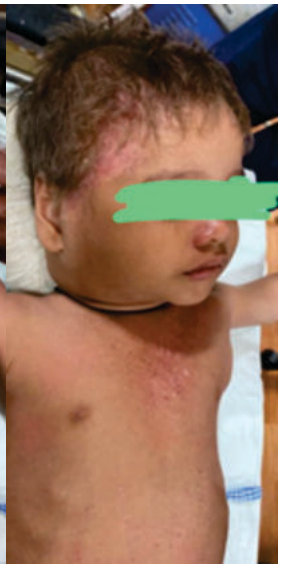
Skin lesions characteristic of LCH can limit the available sites for hemodynamic monitoring, necessitating careful consideration of alternative monitoring locations. This limitation highlights the importance of adaptability in selecting appropriate monitoring techniques tailored to the individual patient.

The utilization of regional nerve block techniques, as demonstrated in our case through landmark-guided erector spinae plane block and fascia iliaca block, can offer significant benefits. Regional anaesthesia not only reduces the overall anaesthetic requirement but also contributes to enhanced postoperative recovery. In the context of LCH, where systemic involvement and potential complications are of concern, minimising the dosage of general anaesthesia becomes particularly valuable.

In conclusion, the anaesthetic management of patients with Langerhans cell histiocytosis demands a comprehensive approach that considers the varied systemic manifestations of the disease. Vigilance in airway management, cautious positioning, meticulous monitoring, and the strategic use of regional techniques collectively contribute to ensuring optimal outcomes in these challenging cases. A collaborative effort among the anaesthesia, surgical, and medical teams is crucial for navigating the complexities associated with LCH and providing safe and effective peri-operative care.



Multiple papulosquamous lesions



Papulosquamous lesions with cervical lympnadenopathy

Declaration Of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient's parent(s) has/have given his/her/their consent for his/her/their child's images and other clinical information to be reported in the journal. The parent(s) understand(s) that their child's names and initials will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

Financial Support And Sponsorship

Nil.

Conflicts Of Interest

There are no conflicts of interest.

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