International Journal of Contemporary Pediatrics
*Shah BK et al. Int J Contemp Pediatr. 2024 Oct;11(10):1450-1452*
http://www.ijpediatrics.com
pISSN 2349-3283 | eISSN 2349-3291

DOI: https://dx.doi.org/10.18203/2349-3291.ijcp20242743

**Case Report**

# Refractory dystonia with intracerebral hematoma after drowning

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**Received:** 15 August 2024
**Accepted:** 12 September 2024

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## ABSTRACT

Drowning in children can have devastating consequences. The risk factors are inadequate supervision, seizure disorder, neurodevelopmental disorder, and cardiovascular disease. The duration of submersion is critical in determining outcomes. The lack of consensus guidelines on pediatric submersion injuries invites difficulty in managing the same. There is a lack of robust evidence on therapeutic hypothermia and, the use of pulmonary surfactants and barbiturates to improve clinical outcomes in such injuries. We present a case of a drowned child in cardiac arrest revived after resuscitation, developing sepsis, seizures, severe dystonia, and intracranial bleeding with hypoxic brain injury requiring vigorous management.

**Keywords:** Child, Drowning, Dystonia, Intracranial hemorrhage

## INTRODUCTION

Drowning stands out as an important contributor to childhood morbidity and mortality.[1] The process of experiencing respiratory impairment from submersion or immersion in liquid is defined as drowning which can be categorized into death, with morbidity or no morbidity.[2] LMIC (Low and middle-income countries) account for a substantial proportion of drowning. The brain suffers from irreversible damage given poor metabolic substrate and asphyxia. The neurological insult is attributed to hypoxemia and hypoperfusion during cardiac arrest. There is often a hemodynamic compromise due to sinus arrhythmia, pulseless electrical activity, and asystole. The early resuscitation of drowning victims leads to favorable outcomes in children.[3]

## CASE REPORT

A two-year-old child presented to casualty in cardiac arrest with an alleged history of drowning. He survived following 30 minutes of CPR (cardio-pulmonary resuscitation) and was shifted to mechanical ventilation in the PICU (pediatric intensive care unit). He required inotropic support, intravenous fluid, and broad-spectrum antibiotics (ceftriaxone, vancomycin). The adrenaline infusion was tapered and stopped after a couple of days. However, given persisting fever spikes, antibiotics were upgraded to meropenem and vancomycin. A lung protective ventilator strategy was used for management. The blood culture was sterile, C-reactive protein was <6 mg/l. However, the airway secretions culture grew Acinetobacter baumannii. Chest X-ray and arterial blood gases were acceptable, but neurological impairment was noted. The hemogram (Table 1) showed hemoglobin of 11.6 g/dl, leukocyte of 17840/cu mm, and platelet of 2.42 lacs/cu mm.

There was no evidence of coagulopathy (INR=1.28) and renal dysfunction (urea 21 mg/dl and creatinine 0.2 mg/dl). There was difficulty in extubating the child from the ventilator given their poor neurological state. He also developed severe dystonia with seizures on day 5 of his illness. Phenytoin was used for controlling seizures at 5 mg/kg/day while dystonia required administration of midazolam infusion at 1 mcg/kg/min. Baclofen was started at 2.5 mg/day but needed escalation up to 30 mg/day and clonidine up to 10 mcg/kg/day. Similarly,

trihexyphenidyl was also added and hiked up to 0.5 mg/kg/day. The dystonia was controlled to a greater extent but not completely. The oxygen support was weaned off by 7 days.

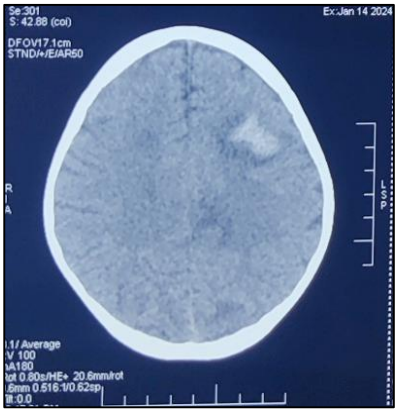

**Figure 1: CECT head revealing left frontal haemorrhage and bilateral diffuse hypoxic changes.**

CECT (Contrast enhanced computed tomography) head revealed a left frontal bleed with bilateral hypoxic changes (Figure 1). A neurosurgical consultation was sought, and advised the use of mannitol, dexamethasone, and antiepileptics. It has been 3 weeks now, seizures have been controlled, and some amount of dystonia is still present. The child is tolerating full feeds via nasogastric tube, is bedridden, and has quadriparesis.

**Table 1: Laboratory investigations.**

| Test | Observed value | Reference value |
|---|---|---|
| **Hemoglobin** | 11.6 g/dl | 11-16 g/dl |
| **Total Leukocyte count** | 17,840 cells/cu mm | 4000-11000 cells/cu mm |
| **Platelets** | 2.42 lakhs cells/cu mm | 1.5-4.5 lakhs cells/ cu mm |
| **Blood urea** | 21 mg/dl | 10-50 mg/dl |
| **Serum Creatinine** | 0.24 mg/dl | 0.6-1.3 mg/dl |
| **Sodium** | 143.9 mmol/l | 136-145 mmol/l |
| **Potassium** | 4.4 mmol/l | 3.5-5 mmol/l |
| **Prothrombin time** | 18 seconds | 11-16 seconds |
| **INR** | 1.2 | 0.8-1.2 |
| **Serum albumin** | 3.4 g/dl | 3.5-5 g/dl |
| **TSB** | 0.15 mg/dl | 0.2-1.2 mg/dl |
| **DSB** | 0.04 mg/dl | 0-0.2 mg/dl |

INR, international normalized ratio; TSB, total serum bilirubin; DSB, direct serum bilirubin

## DISCUSSION

The brain suffers irreversible damage within a couple of minutes of ischemia affecting the cerebral cortex, hippocampus, and caudate nucleus causing injury from selective neuronal necrosis to infarction.[4] The intracranial bleed has not been studied as an association or outcome after drowning as presented in our case. The management of this child required extensive effort in terms of providing cardio-respiratory care and dystonia management. Drowning requiring cardio-pulmonary resuscitation similar to our case has a worse prognosis.[5] There is a lack of experience and clinical expertise in performing stereotactic surgeries and deep brain stimulation targeting globus pallidus, thalamus, and subthalamus for controlling severe dystonia in our country. Moreover, there are intolerability and adverse effects with oral drugs making it difficult to control dystonia.

Aspiration of water (both fresh water and saltwater) dilutes lung surfactant and leads to acute lung injury. There is difficulty in differentiating colonization and infection from the organism isolated from airway secretions as in our case. Lung surfactant therapy as evidenced by a few case reports could not be tried because of financial constraints, and MRI brain also could not be done in view of poor financial status. There is sepsis due to the entry of infected water into the lungs. The antibiotics used in the above case broadly covered gram-positive, negative organisms including Acinetobacter baumannii. Therapeutic hypothermia which has shown benefits in outcome by various trials could not be initiated due to resource-limited setting.[6]

About 95% of children survived neurologically normally in a 2-year follow-up study done by Pearson et al.[7] Similarly, four out of 30 resuscitated drowned children where 17 were survivors, suffered severe neurologic impairment in a prospective study by Kruus et al.[8] There are no studies demonstrating the prevalence and risk factors for intracranial hemorrhage in a drowned child. Good quality evidence for the use of glucocorticoids, surfactants, and barbiturates combined with therapeutic hypothermia in drowning is lacking.

## CONCLUSION

Drowning can lead to cardiac arrhythmia, respiratory arrest with refractory sepsis, and hypoxic brain injury with sequelae. The management should be aggressive in providing adequate neurological and cardiopulmonary support with a multi-disciplinary approach. The index case can help us broaden our knowledge in understanding morbidities associated with drowning survivors and plan effective therapies in maintaining the quality of life for the patients. There is a need for a multicentre paediatric study to formulate standard guidelines for survivors of drowning.

*Funding: No funding sources*
*Conflict of interest: None declared*
*Ethical approval: Not required*

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**Cite this article as:** Shah BK, Kumari N, Singh AK. Refractory dystonia with intracerebral hematoma after drowning. Int J Contemp Pediatr 2024;11:1450-2.