A 7 year-old female with a traumatic cervical spine injury sustained in a motor vehicle collision underwent emergent cervical spine fusion. Her recovery from surgery was complicated by persistent weakness of the left arm that was attributed to a brachial plexus injury. Her cognition and motor function of the right arm and bilateral lower extremities were intact. On the fourth postoperative day she had two generalized seizures that consisted of right-sided jerking and eye deviation to the right with loss of consciousness. The seizures were followed by episodes of hypertension and bradycardia. An MRI was performed to evaluate for hemorrhage, ischemic stroke or progressive cervical dislocation. The images showed diffuse changes of the frontal, parietal and occipital regions bilaterally consistent with posterior reversible encephalopathy syndrome (PRES).

Her seizures were managed with levetiracetam and lorazepam. Her hypertension was controlled with a nicotine infusion followed by oral amiodipine. She had no recurrent seizures or mental status changes. She was dismissed from the hospital on levetiracetam and as needed lisinopril for blood pressure control. A follow-up MRI at 3 months following the episode of PRES revealed complete resolution of the diffuse changes in the frontal, parietal and occipital regions bilaterally. She is currently being weaned off levetiracetam.

A literature review revealed that neither traumatic cervical injury nor traumatic brain injury have been previously described to lead to the development of PRES in children. This etiology should be considered in the differential diagnosis of neurological sequelae of traumatic central nervous system injury in children.

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Background

A literature review of pediatric case reports, case series and articles describing PRES was performed from 1996 through 2012. Studies were included if they contained any pediatric patients (age 0-18). 329 patients from 104 articles were included. Patients were not included if the case description did not contain detail regarding predisposing factors, presenting symptoms, treatment and recovery. The cause was considered chemotherapy-related if the patient received a chemotherapeutic agent as treatment for a hematologic or oncologic malignancy or autoimmune disorder. Recovery data was available for 87.5% of cases (288/329).

Methods

Results

• Three hundred twenty-nine patients were included from 104 case reports or case series. Average age of pediatric patients with PRES was 3 years (0.1-18 years), with male gender predominance (51.7%, 165/329) with gender data. PRES was associated with the administration of chemotherapy in 51.7% of cases reported in the literature (170/329). The most common underlying disease process was hematologic (41.3%, 130/329), which included hematologic malignancy and hemoglobin abnormality (sickle cell anemia). Renal disease was the next most common (38.6%, 127/329) and included chronic renal disease, nephrotic syndrome and acute glomerulonephritis.

Discussion

The exact pathophysiology of PRES is currently unknown; however, understanding of PRES has evolved since its description in 1964 by Hinchey in particular in patients with hypertension. The underlying pathology appears to be development of vasogenic edema. Three primary pathophysiologic mechanisms have been described in the literature. First, hypertension may occur secondary to hypertension autoregulation. Second, medications which cause decreased blood pressure exceed the autoregulation curve, resulting in breakdown of the blood-brain barrier. Third, medications inhibit vasoconstriction which may lead to hypotension with resultant parenchymal ischemic insults that result in edema. Case series have noted edema tends to be localized in watershed areas, consistent with hypertension and ischemic injury. Third, medications (especially the calcineurin inhibitors) are known to cause direct injury to vascular endothelium leading to disruption of the blood brain barrier and vasogenic edema in many patients. A combination of the above may predispose development of PRES.

Conclusion

We present the first documented case of PRES after traumatic cervical spine injury. Even though this appears to be an uncommon sequelae of central nervous system trauma, PRES needs to be included in the differential diagnosis of unexplained neurological findings (seizures, mental status change, headache or vision disturbances) during recovery.

References