Etiologies of secondary craniosynostosis include: metabolic (rickets, hyperthyroidism…), toxic (drugs such as phenytoin, valproate, methotrexate…), hematologic (sickle cell, thalassemia…) and structural (lack of brain growth due to e.g. microcephaly, lissencephaly, micropolygyria…). CSO is rarely associated with hydrocephalus (HCP) [1].

Craniosynostosis following placement of a ventriculoperitoneal shunt for hydrocephalus has been sporadically described. The purpose of an investigation by Bryant et al. was to determine the general risk of developing craniosynostosis in this patient population.

The authors retrospectively reviewed records and radiographs of infants who underwent ventriculoperitoneal shunt placement for hydrocephalus from 2006 to 2012. Recorded variables included date of shunt placement, demographics, comorbidities, cause of hydrocephalus, shunt type, and the number of shunt revisions. Axial computed tomographic images obtained before and immediately after shunt placement and 2 to 4 years after shunt placement were evaluated by a panel of clinicians for evidence of craniosynostosis. Patients with preshunt craniosynostosis, craniosynostosis syndromes, or poor-quality computed tomographic images were excluded. Data were analyzed using STATA Version 15.1 statistical software.

One hundred twenty-five patients (69 male and 56 female patients) were included. The average age at ventriculoperitoneal shunt placement was 2.3 ± 2.58 months. Sixty-one patients (48.8 percent) developed craniosynostosis at a median of 26 months after shunt placement. Of these, 28 patients fused one suture; the majority involved the sagittal suture (n = 25). Thirty-three patients fused multiple sutures; the most common were the coronal (n = 32) and the sagittal (n = 30) sutures. Multivariable logistic regression identified older age at shunt placement and more shunt revisions as independent predictors of craniosynostosis. Shunt valve type was not significant.

Craniosynostosis developed in nearly half of infants who underwent ventriculoperitoneal shunt placement for hydrocephalus. The sagittal suture was most commonly involved. The effect of suture fusion on subsequent cranial growth, shunt failure, or the development of intracranial pressure is unclear [2].

Secondary craniosynostosis occurs in relation to a variety of causes:

endocrine disorders hyperthyroidism hypophosphatemia vitamin D deficiency hypercalcemia hematologic disorders causing bone marrow hyperplasia sickle cell thalassemia inadequate brain growth microcephaly shunted hydrocephalus Shunt-induced craniosynostosis is a rare complication of ventricular shunting for hydrocephalus in pediatric patients. Although the exact pathophysiology of this form of secondary craniosynostosis is not well understood, the current understanding is that persistent drainage of the ventricular shunt causes decreased dural tension, resulting in decreased expansile force on the cranium and premature sutural fusion. Due to the low incidence of this complication, there is no consensus on the ideal treatment for shunt-induced craniosynostosis. In recent years, distraction osteogenesis has been employed with greater frequency, as it is felt to counter the fundamental problem of decreased expansile force on the cranium. However, in a patient with a ventricular shunt, placement of external hardware in close proximity to the shunt could cause significant morbidity due to the increased risk of shunt infection.

Yan et al. presented the management of a patient with shunt-induced craniosynostosis who continued to be shunt-dependent. We chose to use fully buried springs to create an expansile force on the

cranium as an alternative to external distractors so as to mitigate the risk of infection. We demonstrate that spring-assisted distraction osteogenesis can be an effective treatment modality for patients with shunt-induced craniosynostosis. This method should be considered in patients with contraindications to external distraction devices, such as ongoing shunt dependency [3].

1)
Golabi M, Edwards MSB, Ousterhout DK. Craniosynostosis and Hydrocephalus. Neurosurgery. 1987; 21:63–67
2)
Bryant JR, Mantilla-Rivas E, Keating RF, Rana MS, Manrique M, Oh AK, Magge SN, Murnick J, Oluigbo CO, Myseros JS, Rogers GF. Craniosynostosis Develops in Half of Infants Treated for Hydrocephalus with a Ventriculoperitoneal Shunt. Plast Reconstr Surg. 2021 Jun 1;147(6):1390-1399. doi: 10.1097/PRS.0000000000007988. PMID: 34019511.
3)
Yan Y, Bacos JT, DiPatri AJ Jr, Gosain AK. Spring-Assisted Distraction Osteogenesis for the Treatment of Shunt-Induced Craniosynostosis [published online ahead of print, 2020 Aug 12]. Cleft Palate Craniofac J. 2020;1055665620947617. doi:10.1177/1055665620947617