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Clinical Letter

Successful Surgical Treatment of Refractory Status Epilepticus in a 12-Day-Old Infant

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Introduction

Medically refractory epilepsy adds to significant morbidity in the infant population and increases the risk for future neurocognitive dysfunction. Epilepsy surgery should be considered an option in young infants who have catastrophic, localizable epilepsy despite the implementation of optimal first-line therapies.[1,2] There is limited literature focusing on epilepsy surgery in young infants, and more reports and multicenter studies are needed.[1-5] This is the youngest reported patient to undergo successful epilepsy surgery with seizure-free and favorable neurological outcome.

Patient description

The infant was born at term to a 26-year-old mother but developed status epilepticus immediately after birth. He moved all his extremities but had reduced tone. His seizures were characterized by right upper and lower extremity shaking and then transitioned to generalized body stiffening with occasional right hemibody clonic jerking. Scalp video-electroencephalography (EEG) showed interictal and ictal epileptiform discharges originating from his left fronto-centro-temporal region (Fig A,B). Magnetic resonance imaging was also suggestive of cortical dysplasia of the left inferior, middle, and superior frontal gyri (Fig D). At age 12 days and weight 3.71 kg, he remained in status epilepticus despite the use of multiple antiepileptics, and decision was made to proceed with one-stage surgery via a left osteoplastic frontotemporoparietal craniotomy. Electrocorticography demonstrated diffuse burst suppression. Corticectomy was performed following the course of the anterior cerebral artery, and the frontal lobe was removed en bloc without any major complications (Fig E). Postoperatively, no focal motor deficits were observed, and patient's tone significantly improved. Postoperative EEG showed no seizures (Fig C). The pathology revealed extensive cortical dysplasia (Fig F). Now one year after the surgery, the infant remains Engel class I.

Discussion

Careful preoperative planning for epilepsy surgery by a multidisciplinary team is imperative. In the child presented here,

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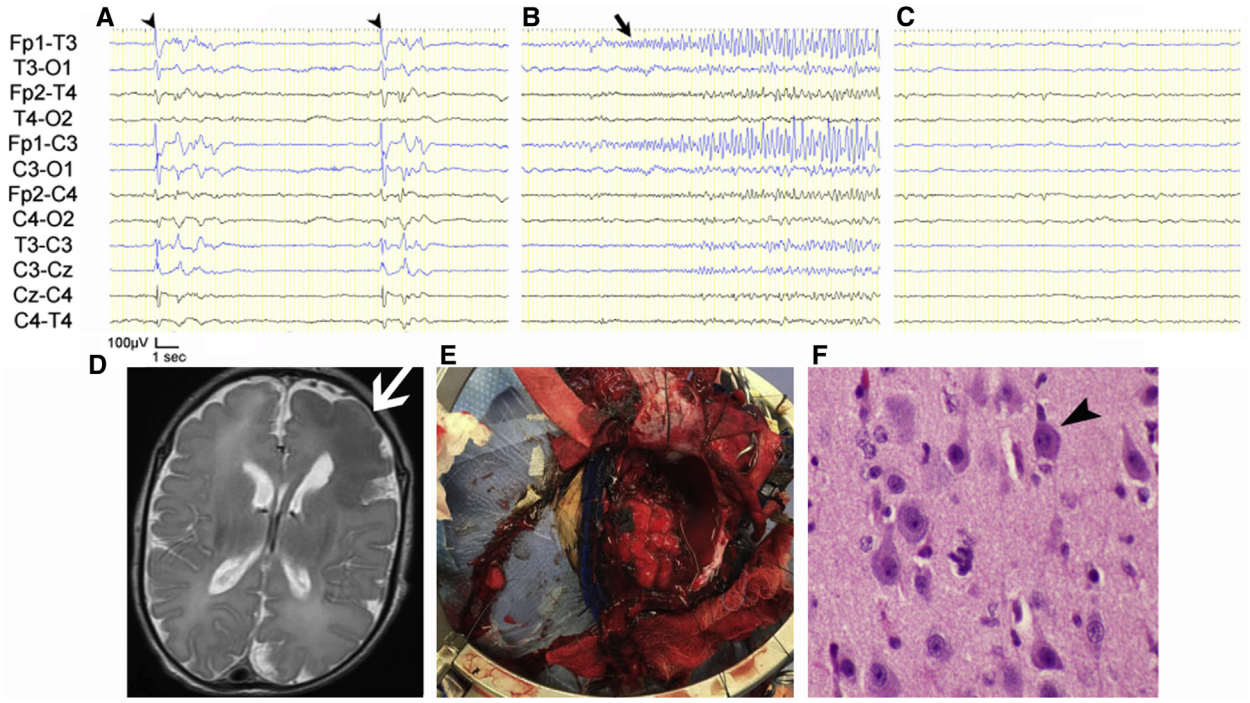

FIGURE. (Two-column image). (A) Video electroencephalograph on day 0 showing high-amplitude irregular spike-and-wave activity (arrowheads) in the left fronto-centro-temporal region during the interictal state. (B) Clinical seizures were associated with sustained rhythmic theta activity (arrow) in the left fronto-centro-temporal region. (C) Four days after the surgery, high-amplitude epileptiform discharges have now disappeared. (D) T2-weighted magnetic resonance imaging showing hypointense gray matter (arrow). (E) Intraoperative gross image post resection. (F) Hematoxylin and eosin stain of the resected cortex with extensive cortical dysplasia with dyslamination and neuronal cytomegaly (arrowhead), 400x magnification. The color version of this figure is available in the online edition.

information obtained from magnetic resonance imaging, seizure semiology, and scalp video-EEG primarily guided the margins for the surgical resection. The posterior margin of the lesion was located anterior to the left precentral sulcus, and expressive language function was thus expected to be reorganized to the right hemisphere, owing to the neuroplasticity in young infants. Moreover, the precentral gyrus and the corresponding precentral artery were preserved, minimizing the chances of a postoperative neurological deficit in our patient.

Intracranial surgery in such young infants carries major challenges when compared with that of the rest of the pediatric population. Thickening of the skull bones, fusion of sutures, closure of the fontanels, growth of the sinuses, myelination, small blood volume, and aberrant anatomy are all important considerations.[6] The patient herein proved to be difficult because of patient's brain tissue friability. Our patient lost 300 mL blood but did not experience hemodynamic instability. Previous reports confirm that significant blood loss is one of the most common risks of epilepsy surgery in young infants.[1-4]

Surgery for intractable epilepsy in young infants can still yield favorable outcomes (Table).[1-5] Kumar et al. have described hemispherotomy performed on an 11-day old infant with hemimegalencephaly; however, this surgery had to be aborted due to hemodynamic instability from significant blood loss and had to be completed when the infant was older.[1] In conclusion, epilepsy surgery in very young infants should be reserved for those with catastrophic epilepsy when first-line options fail. Our patient remains the youngest infant to be successfully treated with epilepsy surgery, hence surgical intervention in young infants is formidable with appropriate planning.

TABLE.
Reports of Epilepsy Surgery in Young Infants

Paper	Youngest Age (Months)	Surgery	Complications	Engel Class
Kumar et al.	.25	AH	Excessive blood loss with instability	II
Duchwony et al.	1	AH	Neurological deficit	I
Gowda et al.	1.5	FL	Blood loss; major vessel territory infarct	N/A
Gonzalez-Martinez et al.	3	AH	Blood loss; neurological deficit	I
Oluigbo et al.	4	EMBO; AH	Excessive blood loss with instability; neurological deficit	I

Abbreviations:
AH = Anatomic hemispherectomy
EMBO = Embolization
FL = Frontal lobectomy
NA = Not applicable.[1-5]
Some of the previously published cases of epilepsy surgery in infants. Only the youngest reported patient in each study is reported (in months). The type of surgery, various complications, and outcome based on Engel classification are also reported.

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