Awake craniotomy in a developmentally delayed blind man with cognitive deficits

Craniotomie éveillée chez un homme aveugle avec un retard de développement et des troubles cognitifs

Mark Burbridge, MD · Mateen Raazi, MBBS

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Abstract

Purpose To describe the complex perioperative considerations and anesthetic management of a cognitively delayed blind adult male who underwent awake craniotomy to remove a left anterior temporal lobe epileptic focus.

Clinical features A 28-yr-old left-handed blind cognitively delayed man was scheduled for awake craniotomy to resect a left anterior temporal lobe epileptic focus due to intractable epilepsy despite multiple medications. His medical history was also significant for retinopathy of prematurity that rendered him legally blind in both eyes and an intracerebral hemorrhage shortly after birth that resulted in a chronic brain injury and developmental delay. His cognitive capacity was comparable with that of an eight year old. Since patient cooperation was the primary concern during the awake electrocorticography phase of surgery, careful assessment of the patient's ability to tolerate the procedure was undertaken. There was extensive planning between surgeons and anesthesiologists, and a patient-specific pharmacological strategy was devised to facilitate surgery. The operation proceeded without

complication, the patient has remained seizure-free since the procedure, and his quality of life has improved dramatically.

Conclusion This case shows that careful patient assessment, effective interdisciplinary communication, and a carefully tailored anesthetic strategy can facilitate an awake craniotomy in a potentially uncooperative adult patient with diminished mental capacity and sensory deficits.

Résumé

Objectif Décrire la complexité des considérations périopératoires et de la prise en charge de l'anesthésie chez un homme aveugle avec retard de développement subissant une craniotomie éveillée pour retirer un foyer épileptique situé au niveau du lobe temporal antérieur gauche.

Éléments cliniques Un homme de 28 ans, aveugle, gaucher et présentant un retard de développement, devait subir une craniotomie éveillée pour réséquer un foyer épileptique au niveau du lobe temporal antérieur gauche causé par une épilepsie réfractaire malgré de nombreux médicaments. Ses antécédents médicaux étaient également importants en raison d'une rétinopathie de prématurité qui l'avait rendu aveugle des deux yeux d'un point de vue légal ainsi que d'une hémorragie intracérébrale survenue peu après la naissance, laquelle a entraîné une lésion cérébrale chronique et un retard de développement. Ses capacités cognitives étaient comparables à celles d'un enfant de huit ans. Étant donné que la préoccupation principale pendant la phase d'électrocorticographie éveillée de la chirurgie était la coopération du patient, une évaluation minutieuse de sa capacité à tolérer l'intervention a été menée. Les chirurgiens et les anesthésiologistes ont effectué une préparation détaillée, et un dosage de médicaments adapté

Author contributions Mark Burbridge performed the literature search for articles used in the discussion and confirmed that this case report had not been described in other publications. The initial manuscripts were written primarily by Dr. Burbridge. He made revisions to the manuscript based on important feedback from the co-author, Mateen Raazi, and prepared the final manuscript for submission. Mateen Raazi reviewed the literature used in the preparation of this manuscript, assisted with writing and reviewing the manuscript, and offered important feedback to the primary author.

M. Burbridge, MD (✉) · M. Raazi, MBBS
Department of Anesthesiology, Perioperative Medicine and Pain Management, Royal University Hospital, University of Saskatchewan, G525-103 Hospital Drive, Saskatoon, SK S7N 0W8, Canada
e-mail: mark.burbridge@usask.ca

au patient a été mis au point afin de faciliter la chirurgie. L'opération s'est déroulée sans complication, le patient n'a pas eu de crise depuis l'intervention, et sa qualité de vie s'est considérablement améliorée.

Conclusion *Ce cas démontre qu'une évaluation méticuleuse du patient, une communication interdisciplinaire efficace et une stratégie soigneusement personnalisée pour l'anesthésie peuvent faciliter une craniotomie éveillée chez un patient adulte potentiellement récalcitrant et présentant des capacités mentales amoindries et des troubles sensoriels.*

Epilepsy affects up to 24% of those who possess significant mental deficits, and neurosurgical resection of epileptogenic foci has been successful for patients who do not obtain adequate seizure control on medications alone.¹ These procedures have usually been performed under general anesthesia.

We describe herein the successful anesthetic management of a cognitively delayed adult male with diminished mental capacity and sensory deficits who underwent an awake craniotomy to resect a left temporal epileptic focus. Preoperatively, comprehensive patient assessment and extensive communication between members of the anesthetic and surgical teams were undertaken. We also describe the carefully tailored anesthetic strategy, including infusions of dexmedetomidine, propofol, and remifentanyl, which facilitated this surgical procedure. Consent was obtained from the patient's legal guardian prior to submission of this article for publication.

Case description

A 28-yr-old left-handed male (height, 173 cm; weight 75.8 kg) was scheduled for an awake left temporal lobectomy to resect an epileptic focus refractory to medical treatment despite multiple medications. The patient had been experiencing seizures since 21 months of age. These seizures were primarily complex-partial but less commonly included grand mal seizures as well as several episodes of status epilepticus. An awake craniotomy was indicated in order to perform electrocorticography due to the close proximity of the epileptic focus to speech areas of the brain. His medical history was also significant for a complicated birth with significant fetal distress and subsequent intracerebral hemorrhage resulting in chronic brain injury and developmental delay. He also suffered from retinopathy of prematurity which rendered him legally blind in both eyes, and he could only vaguely discriminate between colours. Formal intelligence testing had never been carried

out, but estimates placed his mental capacity at that of an eight year old. Surgically, he had strabismus repair when he was three years old without complications under general anesthesia. He was taking the following medications for seizure control: levetiracetam 750 mg *po* bid, topiramate 150 mg *po* bid, and clobazam 40 mg *po* tid. He had no known drug allergies. Physical examination revealed no significant abnormalities of the cardiac or respiratory systems. His neurological examination was also grossly normal, except for bilateral blindness and the described cognitive deficits. The airway examination did not indicate any predicted difficulties for tracheal intubation.

A preoperative consultation with the anesthesiology team took place one week prior to surgery. The patient and his mother, with whom he had an excellent rapport, were present for this interview. The primary objective was to explain how the surgery would be carried out, with emphasis on the "awake" phase involving the patient's tasks to engage in conversation, move parts of his body on command, and repeat words while the surgical procedure was occurring. Secondly, the consultation provided an opportunity to determine if the patient would be able to tolerate surgery based on his level of understanding of the procedure and his level of anxiety. The patient showed an understanding of his need for surgery and how it would be carried out, and he had a strong motivation to proceed with surgery that was cautiously shared by his mother. The next day, the neurosurgical and anesthetic teams met to discuss the case at length, specifically regarding the prospect of the patient suddenly becoming uncooperative during the awake electrocorticography phase. In such circumstances, it was mutually decided that the patient's sedation level would be immediately deepened and surgery would be terminated. This plan was subsequently accepted by the patient and his mother.

The patient's seizure medication was continued to the morning of surgery. The patient was transferred to the operating room with the aid of two nurses. Standard monitors were applied, and oxygen was administered by end-tidal CO₂ sampling nasal cannula at a rate of 3 L·min⁻¹. Two intravenous lines, an 18G on the left forearm and a 20G on the right forearm, were established after four attempts, which the patient tolerated well with the aid of 0.5 mL of subcutaneous 1% lidocaine infiltration and reassurance from the operating room staff. The following intravenous infusions were then started on separate infusion pumps through the 18G: dexmedetomidine 0.5 µg·kg⁻¹·hr⁻¹, remifentanyl 0.05 µg·kg⁻¹·min⁻¹, and propofol 25 µg·kg⁻¹·min⁻¹. After approximately ten minutes, the patient was sufficiently sedated such that an arterial line was placed in the left radial artery on the first attempt with no reaction from the patient. The surgeon infiltrated 0.25% bupivacaine 10 mL without epinephrine

into the patient's scalp at the Mayfield frame pin sites, and the Mayfield frame was applied uneventfully with no change in the patient's vital signs. Next, 0.25% bupivacaine 10 mL without epinephrine was then infiltrated sequentially as skin and then bone were penetrated with no visible discomfort on the part of the patient; at this stage the patient was not able to respond to verbal commands. Upon instruction from the neurosurgeon that electrocorticography would commence, the infusions were adjusted to dexmedetomidine $0.3 \mu\text{g}\cdot\text{kg}^{-1}\cdot\text{hr}^{-1}$, remifentanyl $0.05 \mu\text{g}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$, and propofol $5 \mu\text{g}\cdot\text{kg}^{-1}\cdot\text{hr}^{-1}$. Approximately ten minutes elapsed before the patient responded verbally to hearing his name. The anesthesiologist explained to the patient in a calm voice that he was in the operating room and that he needed to stay calm during electrocorticography. The patient remained fully cooperative and calm and composed during this phase of the procedure.

Cortical mapping proceeded with the patient clearly counting backwards and forwards and responding appropriately to questions, such as "What is your name?" and "Tell me where you are?" Once this phase was completed, the sedative infusions were re-adjusted to their respective pre-electrocorticography rates and the patient did not respond behaviourally or physiologically to any further surgical stimuli. The remainder of the operation was uneventful. Spontaneous ventilation at a rate of $10\text{--}14 \text{ breaths}\cdot\text{min}^{-1}$ was obtained and hemodynamic status remained stable at all times. No airway devices were required at any stage. At the conclusion of surgery, with skin closure and removal of the Mayfield frame, remifentanyl and propofol infusions were discontinued and the dexmedetomidine infusion was decreased to $0.3 \mu\text{g}\cdot\text{kg}^{-1}\cdot\text{hr}^{-1}$; the patient regained consciousness approximately ten minutes later. After completion of a neurological examination by the surgical team, the dexmedetomidine infusion was stopped and the patient was transferred to the recovery room awake, alert, warm, and comfortable. No postoperative complications occurred. On postoperative day one, the patient reported that he had good recall of the procedure; he could remember a number of the questions that were asked of him, but he was not disturbed by this experience in any way. The patient has been seizure-free since the operation, and his quality of life has improved dramatically.

Discussion

Intractable epilepsy is one of several indications for awake craniotomy. Additional indications include surgery for aneurysms, arteriovenous malformations, deep brain nerve stimulator placement, and eloquent cortex and supratentorial tumour resections.² Anesthetic goals for this operation

include facilitating patient cooperation, adequate sedation or anesthesia, anxiolysis, analgesia, and cardiorespiratory stability. These requirements must be synchronized with the specific stage of surgery without interfering with neurological or cognitive testing.³

Awake craniotomy was first performed by Horsley in 1884 and subsequently by the renowned Canadian neurosurgeon, Wilder Penfield.⁴ Despite the long history, there is no uniform anesthetic technique.⁵ Two principle methods are employed, however, i.e., asleep-awake-asleep, and sedate-asleep-sedate.² In the first method, general anesthesia is induced and a laryngeal mask airway device or endotracheal tube is used to protect the airway and enable positive pressure ventilation until the cortical mapping stage, at which point the patient is awakened and the airway device removed to enable verbalization. When electrocorticography is finished, the airway device is again inserted after general anesthesia is established and the surgery is then completed. In contrast, the second approach is accomplished with a varying degree of sedation preceding electrocorticography with the patient breathing spontaneously. Sedation is then stopped to enable patient communication during the mapping phase, and upon completion, the level of sedation is deepened.² Numerous pharmacological agents have been used in both techniques and can be comprised of combinations of midazolam, fentanyl, remifentanyl, propofol, and dexmedetomidine.⁵ Increasingly, institutional protocols are providing safe, effective, efficient, and reliable delivery of anesthetic care to carefully selected homogenous patient groups.⁶

Although awake craniotomy is generally well tolerated, especially when the patient has been adequately educated about the procedure, Whittle³ found that 29% of patients reported experiencing anxiety; 14% felt fear during the procedure, and 20% experienced pain that worsened with manipulation of surgical fixation device pins and skin incision. In addition, in a qualitative study, Khu⁷ reported that recall of intraoperative events was highly variable, and when remembered, recalled events were generally not associated with negative emotion. A delicate anesthetic balance must therefore be achieved regardless of the stage of surgery. Prior to electrocorticography, the patient must be sufficiently comfortable to tolerate the craniotomy, but oversedation must be avoided at the risk of airway obstruction, cardiorespiratory compromise, and aspiration. Conversely, during the awake stage, the patient must not be overly anxious or fearful yet comfortable to ensure adequate cooperation without resisting the urge to move due to discomfort.⁸

Despite the precarious nature of anesthetic care for this procedure, it has been successfully carried out in the past in potentially uncooperative patients. Several cases illustrate the feasibility of this approach in patients as young as eight

years old; however, there is a paucity of case reports regarding awake craniotomy in cognitively delayed adults. A comparable case was described by Ard *et al.*⁹ who realized success in using a primarily dexmedetomidine-based anesthetic in a 12-yr-old girl who was reported to be in the “low average range” of intelligence.

In our case of a cognitively delayed patient, the major concern was intraoperative cooperation during the awake electrocorticography phase of the surgery. There are no accepted criteria to screen cognitively delayed adults for this procedure or any similar procedures. Tobias and Jimenez¹⁰ comment on the selection criteria in pediatric patients undergoing awake craniotomy. They suggest that patient selection and preparation are the most important determinants for a successful surgery and that age and level of maturity are the most important factors in gauging how the patient will tolerate the awake portion of the operation. Factors that led us to believe our patient would be an acceptable candidate during our preoperative interview included his overwhelming desire to have the surgery, his understanding that some of the surgery would be performed while he was awake, his insistence that he would be able to stay calm during the awake phase, his generally calm demeanor during the interview, and his ability to show a general understanding of what was being said to him and to respond appropriately. It was also reassuring that he seemed to understand the logic behind the need to be awake, which shed light on his mental capacity. In a study on adult patients preparing for an awake craniotomy for glioma surgery, Jaaskelainen¹¹ found that, although most patients are first horrified at the idea of awake surgery, almost all patients accepted the idea when they realize that the objective is to avoid unnecessary neurological deficits. Our patient revealed a similar understanding to his cognitively intact adult counterparts that showed a sufficiently mature thought process that we considered would allow him to tolerate the procedure. This opinion was weighed against his mother reporting that her son not infrequently became agitated when scared or anxious and that the proposed operation would be by far the most stressful event he had ever encountered. For this reason, she could not be certain of his full cooperation. After the patient was assessed by both surgical and anesthetic team members, it was decided that he did seem to have the maturity level needed to tolerate the procedure, but if at any time he became sufficiently uncooperative, the patient would be re-sedated and the surgery would be terminated. The patient and his mother consented to this approach.

An effective pharmacological strategy was vital to ensure optimal cooperation given the uncertainty about how well the patient would tolerate the operation. The primary drug employed was dexmedetomidine. This drug is a highly selective alpha-2 adrenergic agonist with a rapid

onset of action, a distribution half-life of six minutes, and a clearance half-life of two hours. It is metabolized in the liver and its metabolites are excreted by the kidneys. Its action is produced by decreasing norepinephrine release primarily at presynaptic neurons in the locus ceruleus in the brainstem, resulting in diminished central nervous system activity.¹² The distinctive feature of this drug is its effective sedative, analgesic, and anti-shivering qualities in combination with an exceptional ability to maintain respiratory drive and patient cooperation.¹³ These favourable qualities were emphasized to surgical colleagues as well as to the patient and his mother preoperatively when considering the patient's ability to tolerate the surgery.

Regarding its use specifically in neurosurgical procedures, its advantageous effects include cooperative sedation, which attenuates hypertension and tachycardia, decreased cerebral blood volume during surgery, optimization of cerebral oxygen supply, neuroprotection, decreased intracranial pressure, and an inability to suppress epileptiform activity on the electroencephalogram. For these reasons, dexmedetomidine has been used with success in awake neurosurgical operations for more than a decade.^{12,13} An infusion of propofol was used to augment the sedative effects of dexmedetomidine, but at a low enough dose to prevent cardiorespiratory depression. Remifentanyl was employed for easy titratability and short-acting analgesia.

This case report describes a successful awake craniotomy in a cognitively delayed adult male with diminished mental capacity and sensory deficits. Careful patient assessment, effective interdisciplinary communication between anesthesia and surgery, and a carefully tailored anesthetic strategy were the most vital components in our approach.

Competing interests None declared.

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