Improvement in Severe Self-Mutilation Following Limbic Leucotomy: A Series of 5 Consecutive Cases

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Background: The efficacy of neurosurgical intervention for self-mutilation behavior associated with severe, intractable psychiatric disorders remains undetermined. We report the effects of limbic leucotomy in 5 consecutive patients with severe self-mutilation behaviors.

Method: After unsolicited referrals from their psychiatrists and careful consideration by the Massachusetts General Hospital Cingulotomy Assessment Committee (MGH-CAC), 5 patients were treated with limbic leucotomy. Their primary DSM-IV psychiatric diagnoses were either obsessive-compulsive disorder or schizoaffective disorder. Comorbid severe, treatment-refractory self-mutilation was an additional target symptom. Outcome was measured by an independent observer using the Clinical Global Improvement, Current Global Psychiatric-Social Status Rating, and DSM-IV Global Assessment of Functioning scales in addition to telephone interviews with patients, families, their psychiatrists, and treatment teams. The mean postoperative follow-up period was 31.5 months.

Results: All measures indicated sustained improvement in 4 of 5 patients. In particular, there was a substantial decrease in self-mutilation behaviors. Postoperative complications were transient in nature, and postoperative compared with preoperative neuropsychological assessments revealed no clinically significant deficits.

Conclusion: In carefully selected patients as described in this report, limbic leucotomy may be an appropriate therapeutic consideration for self-mutilation associated with severe, intractable psychiatric disorders.

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Currently, the accepted approach for chronic, severe, treatment-refractory psychiatric diseases involves a combination of psychopharmacologic medications, psychological interventions, and electroconvulsive therapy (ECT) when indicated. Despite modern treatment methods, a significant number of patients fail to respond adequately and remain severely disabled.[1–3] Many of these patients must live in institutions and are unable to enjoy most normal daily functions. Some of these patients engage in self-mutilation, increasing their likelihood of premature morbidity and mortality.[4] Neurosurgical intervention may be considered as a treatment of last resort in these desperate circumstances.[1–3]

We report 5 severely disabled, treatment-refractory patients with self-mutilation who were treated by limbic leucotomy at the Massachusetts General Hospital, Boston. Self-injurious behaviors included swallowing foreign objects, which required multiple endoscopic retrievals; forced proptosis, which caused blindness; head-banging, which caused loss of consciousness, detached retinas, and subdural hematoma; and self-inflicted third-degree burns. The results of limbic leucotomy over a mean postoperative follow-up period of 31.5 months (range, 16–47 months) are presented.

METHOD

After a mean disease duration of 23.6 years (range, 13–29 years), 5 patients (mean age = 35 years; range, 24–46 years) were treated at Massachusetts General Hos-

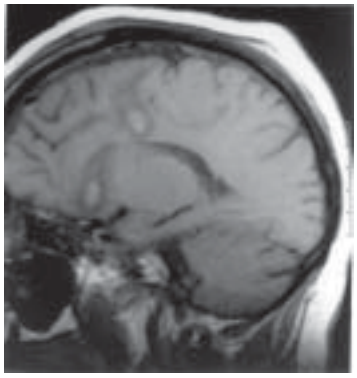
pital with limbic leucotomy in an attempt to prevent or decrease further serious self-injury. The Massachusetts General Hospital Cingulotomy Assessment Committee (MGH-CAC) represents a national referral center for patients with severe, treatment-refractory obsessive-compulsive and affective disorders.[2,3] Patients and/or their guardians gave their informed consent after the procedure and possible side effects were fully explained.

All procedures were performed using the technique described by Kelly,[5] with bilateral radiofrequency thermocoagulation lesions placed in the anterior cingulate and posterior medial orbitofrontal regions. After placement of the magnetic resonance imaging (MRI)-compatible Cosman Roberts Wells stereotactic frame (Radionics, Inc., Burlington, Mass.), sagittal T-1 weighted images (TR300, TE17) were obtained on a GE Signa 1.5 Tesla unit (GE Medical Systems, Waukesha, Wis.) to identify the cingulate gyri and the subcaudate regions bilaterally and to approximate the location of burr holes and planned electrode trajectory. Oblique coronal images (4 mm thickness and 1 mm intervals) were obtained parallel to the proposed trajectory and target coordinates calculated for a point in the anterior cingulate gyrus 1 to 2.5 mm posterior to the tip of the frontal horns, 7 mm from the midline, and 1 mm above the roof of the ventricles bilaterally. Additional target coordinates in the cingulate bundle were calculated at the same anteroposterior and vertical positions, but 14 mm from the midline bilaterally. In the limbic leucotomy procedure, oblique T-1 axial images (3 mm thickness) were also used to target the subcaudate region 7 mm and 14 mm from the midline and 6 mm above the planum sphenoidale. Radiofrequency thermocoagulation lesions were created by inserting an electrode with a 10-mm uninsulated tip to the target coordinates then heating it to 85ºC for 90 seconds. The resulting lesions in the anterior cingulate region measured approximately 2 cm in vertical height and 20 mm in diameter. In the subcaudate region, the resulting lesions were approximately 15 mm in height, 10 mm in depth, and 20 mm in width. Postoperative MRI scans were performed to document adequate lesion placement (Figures 1A–C). The size of the lesions, when measured by repeat MRI several months later, was reduced 30% to 40% in all 3 dimensions.[6]

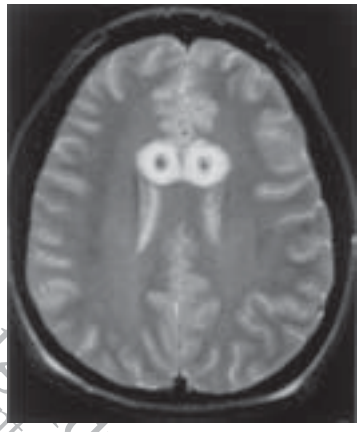
Formal instruments used to measure outcome have been used in previously reported studies assessing patient's response to limbic system surgery[1,2,6–9] and were used in our study. They include the Clinical Global Improvement (CGI) Scale, a 7-item scale ranging from 1 = very much improved to 7 = very much worse, and the Current Global Psychiatric-Social Status (CGPS) Scale,[10] a 7-item scale that categorizes the patient's functional status as S = completed suicide, 0 = worsening of illness, 1 = no change to 5 = normal functioning (Table 1). The DSM-IV Global Assessment of Functioning (GAF) Scale[11] was also employed (Table 2). It considers psychological, social, and

Figure 1. Acute Postoperative (< 48 hours) Magnetic Resonance Imaging (MRI) Scans Documenting Placement of Limbic Leucotomy Lesions

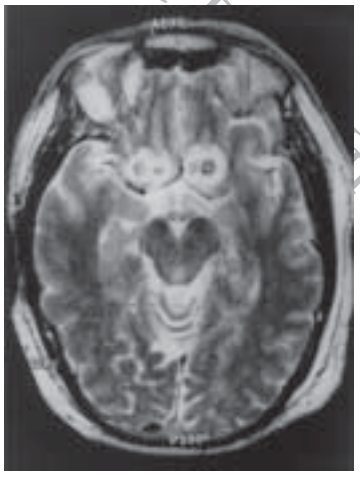
1A. T-1–Weighted Sagittal Image Showing Lesions in the Anterior Cingulate Gyrus and Posterior Media Orbitofrontal Regions[a]



1B. T-1–Weighted Axial Image Showing Extent of Lesions in the Cingulate Gyrus[a]



1C. T-2–Weighted Axial Image Showing Extent of Lesions in the Posterior Medial Orbitofrontal Gyri[a]



[a]The size of the lesions, when measured by repeat MRI scans several months later, is reduced 30% to 40% in all 3 dimensions.[6]

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Table 1. Post–Limbic Leucotomy Function Ratings Over Time in 5 Patients With Self-Mutilation Behavior[a]

Patient/Months	CGI Score[b]	CGPS Score[c]
Patient A		
25 mo	2	3
36 mo	2	3
47 mo	2/3	3
Patient B		
11 mo	2	2
22 mo	2	3
33 mo	2/3	3
Patient C		
17 mo	2	3
28 mo	2	3
40 mo	3/4	1
Patient D		
14 mo	3	2
24 mo	3	2
36 mo	2/3	2
Patient E		
4 mo	2/3	2
16 mo	2/3	2

[a]Patients and families tended to give higher scores, suggesting greater improvement, than their local psychiatrists and treatment team or members of the Massachusetts General Hospital Cingulotomy Assessment Committee. The more cautious estimates of benefit were used.
[b]Clinical Global Improvement (CGI) scores: 2 = much improved, 3 = minimally improved, 4 = unchanged.
[c]Current Global Psychiatric-Social Status (CGPS) scores: 1 = patient shows no change, neither improvement nor decompensation, 2 = patient shows slight improvement and better response to treatment, but still requires intensive care and is unable to work, 3 = patient is considerably improved over preoperative state, no longer critically ill or institutionalized, usually working to some extent, but still displays many serious problems or suffers periodic recurrence of disabling symptoms, requiring continuing psychiatric supervision.

Table 2. Preoperative Versus Postoperative Limbic Leucotomy Global Assessment of Functioning (GAF) Scores[a]

Patient/Time	GAF Score
Patient A	
Preoperative	5
Postoperative	
36 mo	65
47 mo	70
Patient B	
Preoperative	10
Postoperative	
22 mo	60
33 mo	60
Patient C	
Preoperative	5
Postoperative	
28 mo	65
40 mo	5
Patient D	
Preoperative	5
Postoperative	
24 mo	35
36 mo	40
Patient E	
Preoperative	5
Postoperative	
4 mo	43
16 mo	45

[a]GAF scores: 1–10 = persistent danger of severely hurting self or others, 91–100 = no symptoms and superior functioning in a wide range of activities.

occupational functioning on a hypothetical continuum of mental health–illness, with scores from 1–10 indicating persistent danger of severely hurting self or others to 91–100 indicating no symptoms and superior functioning in a wide range of activities. Other scales such as the Beck Depression Inventory (BDI)[12] and Yale-Brown Obsessive Compulsive Scale (Y-BOCS)[13] were given preoperatively and postoperatively when relevant. A psychiatrist not involved in the selection of patients and unknown to the patient, family, and referring psychiatrist conducted an open-ended interview that assessed the patient's daily life, psychiatric status, level of functioning, and possible side effects due to surgery. If postoperative emotional blunting and apathy were not spontaneously mentioned, specific questions regarding these symptoms were directly asked. Preoperative and postoperative neuropsychological assessments were done when possible.

CASE PRESENTATIONS

Patient A

Ms. A was referred for severe, treatment-refractory, self-mutilation behaviors including swallowing foreign objects, which required over 500 endoscopic procedures for removal. Ten abdominal laparotomies including a partial gastrectomy, 3 retropharyngeal abscesses, and 1 esophageal perforation were also documented. Other behaviors included wrist-cutting and head-banging.

She was physically and sexually abused until adolescence and had made several serious suicide attempts. Her first psychiatric hospitalization occurred at the age of 13 years. She was permanently institutionalized at the age of 15. Assaultive behavior toward patients and staff happened repeatedly, requiring 1-to-1 supervision and physical restraints. DSM-IV psychiatric diagnoses included schizoaffective disorder, posttraumatic stress disorder, pervasive developmental disorder, and borderline personality disorder. Family history revealed alcoholism, maternal depression with a history of suicide attempts, and antisocial personality disorders in all 3 siblings. Multiple medication trials were discontinued due to toxicity or lack of benefit. Twenty bilateral ECT and prolonged individual, cognitive/behavioral, and milieu therapies were attempted without success.

When Ms. A was 23 years old, 13 years after the onset of self-mutilation behaviors, bilateral cingulotomy was performed. Transient confusion, headache, ataxia, and lethargy resolved within the first week. Her mood improved and anxiety level diminished after 1 month. She stopped swallowing foreign objects After 2 months, she was permitted to leave the ward by herself. After 9 months, she experienced a relapse of severe depression and resumed

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her preoperative baseline of all prior aberrant behaviors. Thirteen months after the bilateral cingulotomy, a limbic leucotomy was performed. Acute postoperative side effects of blunted affect and urinary incontinence resolved within 1 month. Three weeks after surgery, she had 1 episode of swallowing a foreign object. Four months after limbic leucotomy, she was given day passes. Eighteen months after limbic leucotomy, she obtained her high school equivalency degree and found a part-time cleaning job. She was transferred to a supervised residential program with an individualized behavioral program.

Three years postoperatively, Ms. A was transferred to a less structured residential program. She now goes shopping with her friends without supervision and sees a psychiatrist on a monthly basis only. During extreme circumstances, on average once every 6 months, she engages in superficial wrist-cutting and self-burning with a cigarette. She has not assaulted anyone since the limbic leucotomy. Postoperative medications were not substantially different from preoperative ones.

At 25, 36, and 47 months postoperatively, her CGI score was stable at 2, i.e., much improved. CGPS measured over the same time intervals yielded a stable score of 3, i.e., considerably improved. Her preoperative GAF score was 5; 36 and 47 months after the operation, it was 65 and 70, respectively.

Due to her psychiatric state, Ms. A was unable to fill out the preoperative BDI; 36 and 47 months after the operation, the BDI scores were 22 and 23. Neuropsychological tests administered before bilateral anterior cingulotomy and 28 months after limbic leucotomy revealed a 15-point improvement in the full Wechsler Adult Intelligence Scale-Revised (WAIS-R) as well as recent memory retention, planning and logic, cognitive flexibility, verbal fluency, picture assembly, and block design. Digit span and working memory were unchanged. Perseveration was diminished but present.[14]

Patient B

Mr. B was referred for intractable, compulsive "eye-popping" (forced proptosis), which began at the age of 10 years. He blinded his right eye 11 years prior to referral due to an estimated 30 episodes of forced proptosis per day. Forced proptosis of his left eye 10 to 30 times per day had diminished his visual acuity to 20/70. He was referred with the hope of preserving the sight in his left eye.

Maternal hypothyroidism during pregnancy, speech therapy for stuttering at age 3, delayed toilet training until age 4, and attention-deficit/hyperactivity disorder diagnosed at age 5 were noted. DSM-IV psychiatric diagnoses included pervasive developmental disorder, obsessive-compulsive disorder (OCD), and generalized anxiety disorder. Compulsions involved checking, ordering, symmetry, and hoarding behaviors. There was no family history of psychiatric illness. Since his only psychiatric hospital admission at the age of 5, he had been under constant home supervision by his father. Multiple medication trials were not helpful. Intense individual cognitive/behavioral therapy for 3 years had decreased eye-popping from an average of 30 to 8 times per day. Given impending blindness in his left eye, limbic leucotomy was deemed justifiable.

When Mr. B was 46 years old, 27 years after the onset of forced proptosis, limbic leucotomy was performed. Postoperative urinary incontinence and apathy resolved after 4 weeks. Improvement in the target symptom began after 2 months. At 6 months, despite his father's unexpected death, diminished anxiety and stabilized left vision were noted. By 13 months, forced proptosis of his intact left eye had ceased. At 33 months, he continued taking the same preoperative medications and remained at home by himself with twice weekly nursing visits. Preoperatively, Mr. B utilized psychiatric services every 1 to 4 weeks. He now sees a psychiatrist every 3 months.

At 11, 22, and 33 postoperative months, his CGI score was stable at 2, while his CGPS score was stable at 3. His preoperative GAF score was 10; at 22 and 33 months, it was stable at 60. His preoperative YBOCS score was 26; 22 and 33 months after operation, it was 8 and 12, respectively. His preoperative BDI score was 17; 22 months postoperatively, it was 8. Preoperative neuropsychology testing, repeated 26 months after operation, demonstrated a nonsignificant 4-point elevation in his WAIS-R score, while language and memory functions remained intact. Mild impairments in attention, working memory, and visuospatial construction were unchanged. Improvements in cogitive flexibility and perseveration as measured by Trails B and Wisconsin Card Sort Testing were seen.[14] When directly questioned, his sister (a physician) and treating physician both endorsed mild enduring postoperative apathy and emotional blunting.

Patient C

Ms. C was referred for treatment-refractory self-mutilation, suicidal behaviors, and homicidal thoughts. She had been continuously institutionalized for the previous 10 years, the majority of time spent in 4-point restraints or with 1-to-1 or 2-to-1 staff monitoring. Self-mutilation behaviors included swallowing objects requiring over 20 endoscopies for retrieval, self-burning with cigarettes, wrist-cutting, and attempting to starve herself to death. Multiple suicide attempts by hanging and self-strangulation requiring intensive care had occurred. Multiple assaults on patients and staff by punching, slashing, and burning them with cigarettes were noted. She had fractured the arm of a staff person and sent several staff members to hospitals for emergency care.

A difficult delivery, low birth weight, and hospitalization over the first 4 months of life were noted. She was adopted at the age of 3 years, did not employ language until age 5, and completed toilet training at age 6. At the

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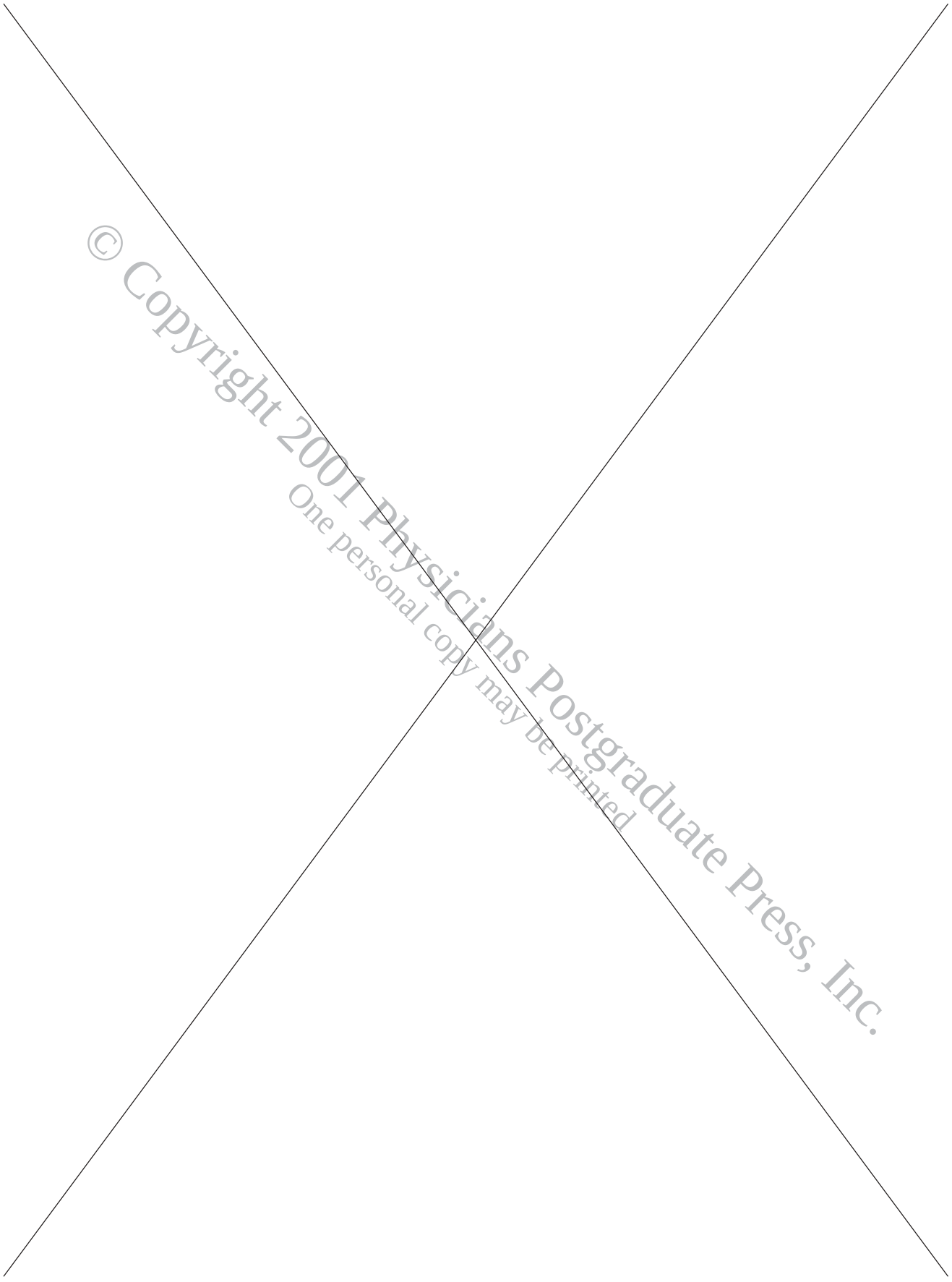

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age of 12, she began abusing multiple substances. Her first psychiatric hospitalization occurred at 15 due to a suicide attempt. DSM-IV psychiatric diagnoses included schizoaffective disorder, anorexia nervosa (past), polysubstance abuse (past), and borderline personality disorder. Multiple psychopharmacologic trials, 30 bilateral ECT treatments, supportive individual and milieu therapies, strict behavior modification, and behavioral management with crisis intervention were employed without benefit.

When Ms. C was 41 years old, 27 years after the onset of self-mutilation and suicidal and assaultive behaviors, limbic leucotomy was performed. Postoperative headache, urinary incontinence, and apathy resolved within 1 month. After 3 weeks, she was noted to be less anxious and more socially engaging. By 2 months, her depression had improved, while suicidal and assaultive behaviors were diminished such that 1-to-1 monitoring was no longer necessary. By 17 months postoperatively, depression and suicidal ideation had resolved, and there were no further assaults. She was transferred to an unlocked but strict behavioral residency program. She had 2 episodes of swallowing foreign objects in the wake of emotional distress without need for endoscopy. No other episodes of self-injury occurred. Social interactions improved, and Ms. C began working in a custodial job with earned income.

At 17 and 28 months postoperatively, her CGI was stable at 2, i.e., much improved, and her CGPS was stable at 3, i.e., considerably improved. Her preoperative GAF score was 5; at 28 months it had increased to 65. Her preoperative BDI score was 38; 28 months postoperatively, it was 9. Ms. C appeared more motivated, wanted to live in an apartment, and desired more gainful employment. Comparing preoperative neuropsychological data with repeat data obtained 16 months after limbic leucotomy, no clinically significant changes were noted in WAIS-R IQ, digit span, working memory, or verbal fluency scores. Substantial improvement in Trails A and B tests was noted.[14] However, when evaluated 40 months postoperatively, she had dramatically relapsed to her preoperative baseline. The CGI score had returned to 4, CGPS score had deteriorated to 1, and GAF score had dropped to 5. Once again, Ms. C often required 1-to-1 monitoring, had become self-injurious, suicidal, and assaultive and remains confined to a locked ward.

Patient D

Ms. D was referred for self-mutilation and ritualistic and other compulsive behaviors. Self-mutilation included violent banging of her head, right hand, and right foot against walls with a subsequent detached retina, festering hand wounds, and fractured right ankle. She had been institutionalized for 8 years at the time of our evaluation. DSM-IV psychiatric diagnoses included Tourette's disorder with onset at age 4, obsessive-compulsive disorder with onset at age 8, and borderline personality disorder. She was verbally and physically assaultive toward other patients and staff. Multiple medication trials and intensive behavioral therapy resulted in no improvement. Family history was positive for OCD, motor tics, and agoraphobia. Preoperative brain MRI was performed under general anesthesia given her refusal to be touched.

When Ms. D was 33 years old, 29 years after the onset of self-mutilation behaviors, a limbic leucotomy was performed. A postoperative fever resolved after 3 days. After 1 month, improvement was noted in obsessions, compulsions, and self-injury. Six months later, her rituals and head-banging returned, resulting in a subdural hematoma without need for evacuation. However, her overall progress continued with fewer compulsions, improved ability to communicate, fewer motor tics, and diminished vocal tic volume. Eight months postoperatively, she was transferred to an unlocked, less structured neurorehabilitation unit where Ms. D continues to reside. At 24 months postoperatively, head-banging persisted, but a behavioral program allowed her to restrict head-banging to 5 minutes out of every hour, which curtailed further significant head injury. She no longer required 1-to-1 staff monitoring and was allowed to walk on the hospital grounds without supervision. Her family was able to take her on day trips. Incidents of serious head-banging, screaming, and kicking the walls or patients were substantially diminished. Her mother stated, "All her behaviors are still there; they come out from time to time, but they are less severe, and she is a much better person now." At 36 months follow-up, Ms. D allowed weekly phlebotomy so that clozapine therapy could be started. She remains on a locked ward but does not require special supervision. Family outings continue.

At 14, 24, and 36 postoperative months, her CGI score was stable at 3, and her CGPS score was stable at 2. Her preoperative GAF score was 5; after 24 months, it had increased to 35; by 36 months it was 40. Preoperative YBOCS score was 40; at 24 postoperative months, it was 34. Postoperative medications remained approximately the same as preoperative ones in addition to clozapine. She was unable to complete neuropsychological tests prior to surgery. However, 33 months after surgery, selected measures of general intelligence, attention, and cognitive flexibility were performed within the average range.[14]

Patient E

Ms. E was referred for treatment-refractory self-mutilation. OCD behaviors started at the age of 5, auditory hallucinations began at age 8, and self-inflicted third-degree burns to her feet occurred at the age of 9. Over the past 11 years she had been hospitalized at 9 different institutions, spending only 61 total days outside of a hospital. Self-mutilation also included head-banging with lacerations requiring multiple sutures to her head, limbs, thorax, and neck; cigarette burns; and lacerations, as well

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as severe finger biting, requiring surgery. A helmet and protective mitts were used to prevent harm. Two-to-one and 1-to-1 monitoring 24 hours per day were required for many months at a time. Six serious suicide attempts were noted. There was no history of assault against other people.

Additional history suggested sexual molestation, an eating disorder that prompted her first psychiatric admission at age 15, hypothyroidism with appropriate treatment, history of alcohol abuse, and borderline personality disorder. Multiple psychopharmacologic trials, 2 courses of ECT, individual biofeedback, cognitive behavioral therapy, and individual, family, and group psychotherapy were not helpful. Dialectical behavioral therapy resulted in mild decrement of self-mutilation. Family history revealed a great aunt with frontal leucotomies for unknown reasons. Both parents suffered from affective disorders.

When Ms. E was 31 years old, 22 years after the onset of self-mutilation and suicidal behaviors, a limbic leucotomy was performed. A postoperative headache cleared after 2 weeks, while flattened affect resolved over 3 months. Sixteen months after the operation, diminished anxiety, improved tolerance of stress, and 1 suicidal gesture were noted by her treating psychiatrist and family. They also reported clinically significantly diminished self-injurious behaviors with only occasional neck scratching and head banging. She remained hospitalized, but required far less 1-to-1 monitoring. At 4 and 16 months postoperatively, her CGI score was variously rated at 2 or 3, while the CGPS score was stable at 2. Her preoperative GAF score was 5; 4 and 16 months after operation, it was 43 and 45, respectively. Her preoperative BDI score was 46; 9 months postoperatively, it was 44.

Neuropsychological assessments performed 3 months preoperatively and repeated 4 months postoperatively showed no substantial change in WAIS-R scores, attention, working memory, Trails A, and Wisconsin Card Sorting Test. Mild postoperative slowing (but without errors) was seen on Trails B.[14]

RESULTS

Postoperatively, 4 of 5 patients demonstrated clinically significantly decreased self-mutilation behaviors. Over a mean postoperative follow-up period of 31.5 months (range, 16–47 months), the CGI and CGPS scores demonstrated stable improvement in 4 patients and no improvement in 1 patient. GAF scores demonstrated an overall mean postoperative increase of greater than 30 points and a mean postoperative increase of greater than 40 points in the 4 responders. Postoperative neuropsychological assessments and measures of daily living activities suggested an absence of major behavioral or cognitive deficits. These are the only patients with self-mutilation whom we have treated by limbic leucotomy.

Deinstitutionalization of Patient A after hospitalization for 9.5 years, preservation of eyesight in Patient B, diminished requirement for supervision and restored ability to go on family outings in Patient D, and greatly reduced need for staff monitoring in Patient E were the most salutary results. The preoperative assaultive behaviors of Patients A and D were also clinically significantly diminished. In addition, Patient D experienced a reduction in her motor tics. The explanation for Patient C's decompensation after 28 months of improvement is not entirely clear but may involve the stress of legal actions taken on her behalf, transfer to another institution, and family turmoil.

DISCUSSION

Self-Injury and Mutilation

Self-injurious behavior is a dramatic but poorly understood phenomenon that refers to the commission of deliberate, direct harm to one's own body with resulting tissue damage.[4,15] Common forms include cutting and burning of skin, banging of head and limbs, picking at wounds, and self-biting. Extreme forms, as illustrated by the 5 patients we report here, include eye enucleation and autoamputation.[4,15] Although patients who repeatedly engage in self-injury are at high risk, such behavior is not considered a suicidal act. Repetitive self-mutilation may be associated with a wide range of maladaptive impulsive behaviors including substance abuse, eating disorder, promiscuity, and physical aggression toward others.[16,17] It is a common reason why families request residential care for their affected relatives.

The literature is complicated by methodological flaws, variable definitions, and the failure at times to differentiate internally from externally directed aggression.[4] The prevalence of superficial or moderate self-injury may range from 400 to 1400 per 100,000 people.[15] It is estimated that at least 1 in 600 adults in the general population wound themselves sufficiently to require hospital treatment.[4] The prevalence rates are generally higher for females than males, although males carry out more violent and destructive acts.[4] In one study, 62% of repetitive self-cutters reported a history of sexual and/or physical abuse.[15] Although self-injury peaks in adolescence and early adulthood, longitudinal studies find that about 90% of self-injurious behaviors are still present 10 years later.[18] The patients' morbidity and mortality rates may be adversely affected.[4,15,18] Self-injury is common in the institutionalized mentally retarded and prison populations as well as outpatients with various intellectual developmental disabilities, eating disorders, autism, and genetic syndromes, including Lesch-Nyhan, Cornelia de Lange, and Rett syndromes, and neuroacanthocytosis.[4,15] Associations with schizophrenia, obsessive-compulsive disorder, bipolar disorder, acute psychosis, borderline personality disorder, Tourette's disorder, and substance abuse have been

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reported.[4,15] Treatment remains largely empirical and often disappointing and may include serotonin reuptake inhibitors, dopamine antagonists, β-blockers, opiate antagonists, anticonvulsants, benzodiazepines, and other GABA-modulating agents.[4,15] Psychological interventions combined with polypharmacy are usually necessary.[4,15,18]

Limbic System Surgery

Following Moniz's report in 1937,[19] the rapid acceptance of psychosurgery was largely attributable to the anguish of so many patients and families for whom available therapies offered unsatisfactory results. Between 1942 and 1954, more than 10,000 patients in England and Wales and more than 18,000 patients in the United States were treated by this method.[3] However, a poor theoretical understanding of psychiatric disease, idiosyncratic and nonspecific psychiatric diagnoses, variable surgical techniques, significant operative complications, and insufficient systematic, controlled follow-up studies imposed further limitations so that psychosurgery remains generally regarded as a treatment of last resort.[3] Over the last 4 decades, stereotactic neurosurgical techniques and brain imaging technology have evolved, making possible the placement of much more restricted and accurate lesions. In addition, improved methods of psychiatric diagnosis and follow-up have allowed more precise characterization and measurement of psychiatric illness.[3] Scientific studies have confirmed the sustained benefits of neurosurgical intervention in obsessive-compulsive disorder, affective disorder, and general anxiety disorder.[2,3,6,8,9]

We prefer the term *limbic system surgery* instead of *psychosurgery* for several reasons. Psychosurgery, which was introduced in the 1940s, encompassed a number of operations that are no longer used and targeted many psychiatric disorders or syndromes for which neurosurgical intervention is no longer considered appropriate.[2,3] The term *limbic system surgery* for psychiatric disorders serves to distinguish between the long-abandoned, blind, freehand neurosurgical "frontal lobotomy" techniques and the current, modern stereotactically guided procedures.[2,3] Finally, *psychosurgery* perpetuates the myth that it is a unique form of surgery. In reality, this procedure is technically similar to other ablative procedures for treatment-refractory neurobiological diseases including seizures, chronic pain, tremors, dystonias, and Parkinson's disease.

While the complex neuroanatomy, neurochemistry, and neurophysiology of psychiatric disease are not completely understood, accumulating evidence suggests that the limbic system is clearly involved. Limbic system surgery involves the direct or indirect perturbation of interconnections between discrete fiber pathways involving structures such as the amygdala, hippocampus, certain thalamic and hypothalamic nuclei, the prefrontal and orbitofrontal cortex, the cingulate gyrus, and cingulum. Two recent studies published by our group[20,21] suggest cingulostriatal perturbation as the putative mechanism underlying the efficacy of bilateral anterior cingulotomy for severe treatment-refractory OCD.

Limbic leucotomy. Since 1962, bilateral anterior cingulotomies for severe, intractable psychiatric illnesses such as OCD, schizophrenia, depression, and anxiety have been performed at Massachusetts General Hospital.[7] In 1987, Ballantine et al.[7] reported that depression or anxiety was improved in 60% of 198 patients followed for at least 8 years after operation. The procedure was deemed less effective in schizophrenia.[1] Additional experience at Massachusetts General Hospital, including 1991 and 1996 retrospective reports[6,8] and a 1995 prospective report of cingulotomy and limbic leucotomy for OCD and/or affective disorders,[9] also demonstrated efficacy. In 1973, Kelly et al.[22] reported that the combination of bilateral stereotactic cingulate and posterior medial orbitofrontal lesions (limbic leucotomy) yielded the best outcome for patients with severe, treatment-refractory psychiatric diseases. In 1976, Kelly's group[23] confirmed an overall response rate of 70% in 66 post–limbic leucotomy patients treated for anxiety, depression, and schizophrenia who were followed over a mean period of 16 months. In 1991, Kelly[5] further reported that 84% of 49 patients who underwent stereotactic limbic leucotomy for intractable OCD were substantially improved over a mean follow-up period of 20 months.

Our decision to proceed with limbic leucotomy in an attempt to prevent or decrease further serious self-injury was based on our experience, Kelly's work, and other case reports. In 1996, we[6] reported that 11 of 31 patients initially treated with bilateral anterior cingulotomy for treatment-refractory OCD and/or affective disorders required second extended lesions for sustained significant improvement. We also confirmed the lack of enduring neurologic, intellectual, personality, or behavioral deficits in this and other series.[1,6–9] Our initial experience with Patient A, who required a limbic leucotomy 13 months after cingulotomy for maximal benefit, also encouraged the use of limbic leucotomy for the subsequent 4 patients. In addition, other single case reports supported the efficacy of limbic leucotomy in self-mutilation. In 1990, Robertson et al.[24] described a 19-year-old man with OCD, Tourette's disorder, and severe self-mutilation behaviors. Six weeks after limbic leucotomy, improvement began. Two years later, self-mutilation behaviors had ceased, tics had improved, no major adverse effects were reported, and he was employed.[24] In 1993, Sawle et al.[25] reported a 45-year-old man with OCD, Tourette's disorder, and self-mutilation behaviors. Nineteen months after limbic leucotomy, no further self-mutilation behaviors had occurred, improvement in OCD and tics were reported, and he lived independently. A 1995 review by Rauch et al.[26] also suggested benefit from limbic leucotomy in patients with Tourette's disorder and self-injurious behaviors.

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Strom-Olsen and Carlisle[27] reported that patients demonstrating the least benefit from neurosurgery were those with a disease duration of 10 years or more. Goktepe and colleagues[28] also noted that those patients with the longest duration of disease and most hospitalizations were the group least likely to benefit from neurosurgery. Given the mean disease duration in our patients before surgery of 23.6 years (range, 14–30 years), it is conceivable that greater benefit might have occurred had limbic leucotomy been performed within the first 10 years of disease onset. Although a series of 5 patients with mixed psychiatric and neurologic diagnoses constitutes a limited experience, our report represents the largest and most extensively studied series of consecutive patients treated for self-mutilation with limbic leucotomy. Prior to limbic leucotomy, no form of psychological, psychopharmacologic, or electroconvulsive treatment had yielded sustained benefit in these patients. While the mean disease duration of 23.6 years and severe, treatment-resistant nature of the illness argue strongly against the spontaneous resolution of symptoms independent of surgical intervention, placebo effect, or wish-fulfillment, these possibilities cannot be entirely excluded.

CONCLUSION

MRI-guided stereotactic limbic leucotomy yielded sustained clinical benefit in 4 of 5 patients with self-mutilation behaviors. Improvement was demonstrated by multiple, clinically validated rating scales. The professional caregivers, families, and patients independently validated and mutually corroborated these results. Assaultive behaviors also declined in 2 of 3 patients, while interpersonal skills, general autonomy, and daily functioning improved in 4 patients. Decreased need for psychiatric attention occurred in 4 patients. Postoperative complications were relatively minor and transient in nature. Other than mild apathy and emotional blunting reported in 1 patient, no major, enduring, postoperative behavioral or cognitive deficits were noted.

We conclude that in carefully selected patients, limbic leucotomy remains a treatment that has potential for benefit. Rigorous prospective studies in specialized centers are needed to verify and expand upon these preliminary results. Finally, the development of reversible, nonablative somatic treatments such as deep brain stimulation and transcranial magnetic stimulation for refractory psychiatric disease may be enhanced by improved understanding of MRI-guided neurosurgical interventions and their consequences.[29,30]

Drug name: clozapine (Clozaril and others).

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