

Hypersexuality Following Septal Injury

David G. Gorman, MD, Jeffrey L. Cummings, MD

● Hypersexuality is an uncommon consequence of brain injury. We report two cases of markedly increased sexual activity following septal damage sustained in the course of placement of ventriculoperitoneal shunts. These two cases, observations in animal experiments, and descriptions of altered behavior in humans with localized brain dysfunction indicate that a circuit involving the septal nuclei has an important role in the mediation of sexual behavior. (*Arch Neurol.* 1992;49:308-310)

Hypersexuality is an infrequently reported behavioral alteration in neurologic illness. Increased sexual behavior has been observed with localized lesions of the frontal lobe,¹ as a component of secondary mania occurring with right hemisphere lesions,¹ with bilateral anterior temporal lobe lesions in the Klüver-Bucy syndrome,² in conjunction with epilepsy³ (particularly in the postictal state or following temporal lobectomy), and as a rare consequence of the treatment of Parkinson's disease with dopaminergic agents.⁴ A unifying explanation of altered sexual behavior in these diverse clinical circumstances has not been formulated.

We recently observed two patients who developed markedly increased sexual behavior following placement of ventriculoperitoneal (VP) shunts for hydrocephalus. In both cases the catheter tip was shown by computed tomography to have penetrated the dorsal septal region. We have not observed any other cases of hypersexuality following VP shunting in the past 10 years, during which a total of 16 patients with VP shunts were studied in a clinic specializing in the evaluation of neurobehavioral disturbances. No instances of septal insertion of a shunt catheter were observed in scans of these patients. Three patients in whom the shunt catheter passed through the dorsal or middle septum pellucidum into the contralateral ventricle avoiding the septal nuclear region evidenced no change in sexual interests or behavior. The relationship between septal injury and hypersexuality in the two pa-

tients described herein indicates that the septal nuclei play an important role in mediating sexual behavior.

REPORT OF CASES

CASE 1.—A 76-year-old right-handed man living in a nursing home was referred for evaluation of inappropriate sexual behavior. He had never been married, courted very little, and had never used coarse or suggestive language. He had a stroke in 1975 with full recovery except for minimal residual gait disturbance. Ten years later he collapsed and was taken to a hospital where he was diagnosed as having normal pressure hydrocephalus, and a VP shunt was placed. He remained unresponsive and was subsequently transferred to another hospital where the shunt was twice revised, improving his mental state.

After the final shunt revision, there were many reports of the patient approaching and fondling female patients. He was noted to crawl into bed with other patients with sexual intent and to use sexually explicit language. He had to be restrained continuously to prevent the sexual behavior. The anomalous behavior had persisted for 3 years at the time of referral.

On examination, the patient was alert and oriented. His digit span was seven digits forward and five in reverse. He could recall three objects after 3 minutes, but remote memory was limited. His insight and judgment were intact and his affect was appropriate. His speech was fluent. He could do simple calculations and constructions. His word list generation was impaired (11 animals named in 1 minute). His performance on serial hand sequences and reciprocal programs was mildly impaired. On neurologic examination, his strength and muscle stretch reflexes were symmetrical; flexion responses were elicited with plantar stimulation. His gait was retropulsive, and he was somewhat dysmetric in all four extremities. An electroencephalogram showed occasional right temporal spikes as well as generalized slowing. A computed tomographic scan (Fig 1) showed no hydrocephalus, but the tip of the catheter was lodged in the septum in the medial aspect of the floor of the lateral ventricles at the junction of the frontal horns.

A series of open-label treatment trials were initiated to control the patient's sexual impropriety. Carbamazepine, haloperidol, propranolol, and diethylstilbesterol were administered in conventional therapeutic doses for a minimum of 1 month each and had no discernible effect on his sexual behavior. He declined shunt revision.

CASE 2.—A 75-year-old right-handed man was referred for evaluation of hypersexuality. In 1983 he had a herpetic rash involving his right eye and forehead. Encephalitis with coma followed, and the patient was treated with adenosine arabinoside. After recovery from the acute phase of the encephalitis, he was transferred to a nursing home. Ventricular enlargement led to placement of a VP shunt.

Prior to the encephalitis, the patient and his wife had weekly sexual relations. In earlier years, he had occasionally expressed an interest in group sex, but when his wife declined he had not

Accepted for publication November 12, 1991.

From the Departments of Neurology (Drs Gorman and Cummings) and Psychiatry and Biobehavioral Sciences (Dr Cummings), UCLA School of Medicine, and the Behavioral Neuroscience Section, Psychiatry Service, West Los Angeles Veterans Affairs Medical Center (Drs Gorman and Cummings), Los Angeles, Calif.

Reprint requests to Neurobehavior Section, Reed Neurological Research Center, 710 Westwood Plaza, Los Angeles, CA 90024 (Dr Cummings).

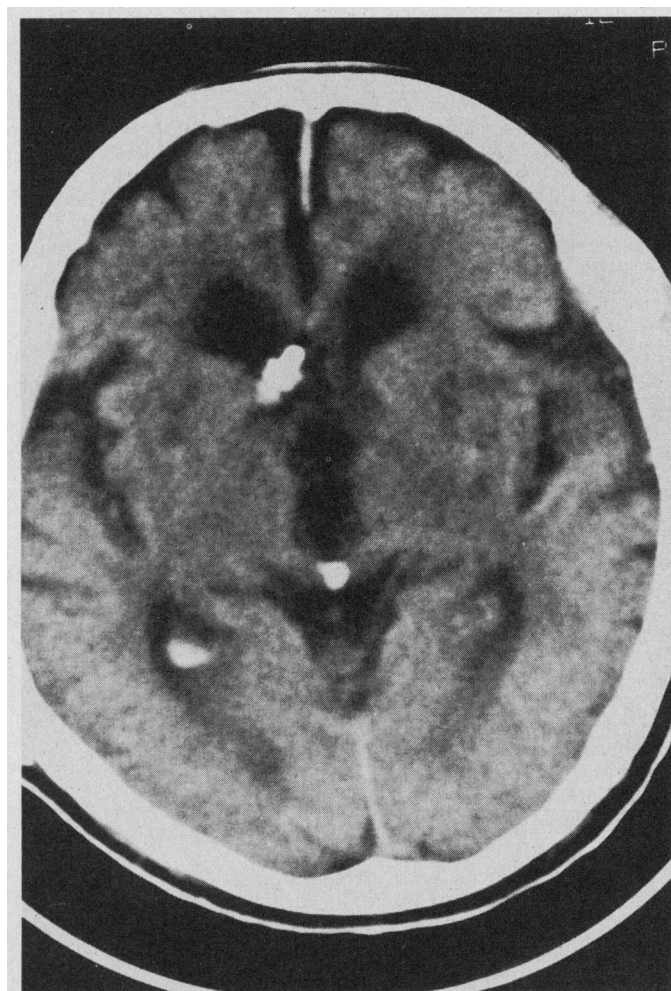


Fig 1.—Case 1. Computed tomographic scan showing catheter tip in septum verum.

insisted, and their sexual relationship was otherwise unremarkable. An increase in sexually motivated activity was apparent after the patient regained consciousness from his encephalitis. He made sexual comments toward women he encountered in the hospital, attempted to fondle the nurses, and masturbated publicly. After discovery of the hydrocephalus and shunt placement, his previously disinhibited sexual behavior was markedly increased and became "disgusting" according to his wife. He became "the man with a thousand hands," attempting to fondle her each time she came within reach. He requested intercourse with her many times each day and also asked that she have sex with other men while he watched, an interest never previously expressed. The excessive sexual interest had been present for 2 years at the time of assessment.

On examination, the patient was alert, and his language, memory, and calculations were intact. He could name only six animals in 1 minute, and he could not interpret proverbs abstractly or copy complex figures. The neurologic examination showed no abnormalities except for mild psychomotor retardation. A computed tomographic scan (Fig 2) revealed hydrocephalus with the tip of the shunt inserted into the midline anterior hypothalamic-septal structures. The patient refused shunt revision. (This case was previously reported by Miller et al.¹)

COMMENT

Markedly increased sexual behavior followed placement of VP shunts in these two patients. In both cases, computed tomography revealed that the shunt catheter tip was lodged in the septum. Several lines of evidence

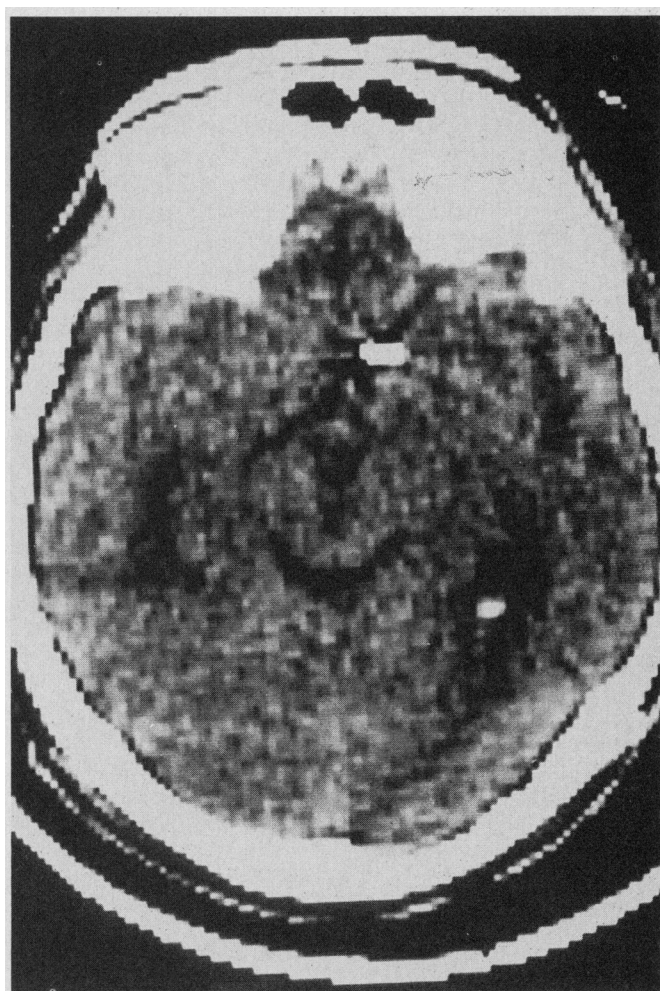


Fig 2.—Case 2. Computed tomographic scan showing catheter tip in septal area.

suggest that the septal injury was responsible for the change in sexual conduct. First, neither patient had evidenced atypical sexual behavior before brain injury. Although patient 2 had disinhibited sexual behavior while in a postencephalitic confusional state prior to shunt placement, there was a dramatic exaggeration of sexual activity in a clear state of consciousness following the shunt insertion. Second, no similar cases of hypersexuality with VP shunts or hydrocephalus in the absence of septal injury have been observed by us or reported by others (to our knowledge). Third, we considered seizures as an alternative explanation for the behavioral changes, but the behavior was not confined to discrete episodes, was not accompanied by impaired consciousness or other evidence of seizures, and was not ameliorated by treatment with carbamazepine (case 1). Fourth, as reviewed below, damage to the septum in animals and humans has previously been associated with hypersexuality.

The septal area has two divisions, the septum pellucidum and the septum verum.⁵ The septum pellucidum is the thin dorsal tissue leaf separating the lateral ventricles and consisting of fiber tracts, glia, and ependymal lining, but no nuclei. The septum verum is ventral to this, between the subcallosal gyrus rostrally and the anterior commissure and anterior hypothalamus caudally. Most authors^{6,7} include as its parts the lateral septal nuclei, the medial septal nuclei and diagonal band of Broca, the pos-

terior septal nuclei, the bed nucleus of the stria terminalis, and the nucleus accumbens septi. The lateral septal nuclei receive their principal input from the hippocampus, and their main output is to the medial septal nuclei. They also receive projections from the ventral tegmental area, the bed nucleus of the stria terminalis, the nucleus accumbens, and the amygdala. The medial septal nuclei and the nuclei of the diagonal bands of Broca have their main input from the lateral septal nuclei; their major output is to the hippocampus via the fornix. The medial septal nuclei and the nuclei of the diagonal bands also have connections with the amygdala, the orbital frontal cortex,⁸ and the hypothalamic areas. The medial septal nuclei send fibers to the ventral tegmental area, locus ceruleus, and raphe nuclei in the brain stem.⁹ The posterior septal nuclei receive input from the hippocampus and direct their output to the habenular nuclei. The stria terminalis starts from the amygdala and ends in the septal region; within this white matter tract there are interstitial or "bed" nuclei that are extensions of the amygdaloid nuclei from the temporal lobe to the septal area.¹⁰ The nucleus accumbens septi, also known as the limbic or ventral striatum, receives its input from the anterior cingulate cortex and projects to the ventral globus pallidus.¹¹

Experiments with animals suggest a role for septal nuclei in mediating sexual behavior. Alterations of sexual activity have been observed with both septal lesions and septal stimulation in a variety of species. Rasmussen et al¹² reported a 400% increase in sexual drive of rats as measured by crossings of an electrical grid to a sex incentive following lesioning of the dorsal septal region. Cats with septal lesions exhibited both an increase in sexual activity and a change to indiscriminate sexuality, mounting rabbits and inanimate objects.¹³ In squirrel monkeys, septal stimulation induced copulatory behavior.¹⁴

Relatively few discrete septal lesions or stimulations have previously been reported in humans. Heath and Fitzjarrell¹⁵ attempted to control seizures in a woman with epilepsy by placing a chemical stimulating catheter directly into the septum. When acetylcholine was introduced into the septal region, the patient became euphoric and experienced sexual orgasm on each of twelve stimulations. At autopsy several years later, the septal position of the cannula was confirmed. Heath¹⁶ also recorded from the septum of patients during sexual intercourse and observed spike-wave activity in the region during orgasm. Sem-Jacobsen¹⁷ performed stimulations in the septal region in two patients and elicited sexual interest or orgasm.

Altered sexual behavior has also been reported with damage to the inferior frontal cortex, injury to the hypothalamus, and bilateral damage to the amygdaloid nuclei.² As described above, each of these areas has major anatomic connections with the septal region, and together they define a circuit of structures mediating sexual behavior. Lesions in any of the regions, particularly if the lesions are bilateral, have a major impact on sexual activity; sexual behavior may be increased or decreased depending on the site of injury. Bilateral lesions of the amygdaloid nuclei produce hypersexuality in the Klüver-Bucy syndrome. Hypothalamic injury, on the other hand, typically results in reduced sexual behavior. Meyers¹⁸ reported impotence and diminished libido following destructive lesions in the medial hypothalamic region made in the course of surgery for parkinsonism, and posterior

hypothalamotomy has been performed to reduce sexual behavior in individuals convicted of sexual misconduct.¹⁹

Dopamine is an important transmitter at several of the synapses of the circuit mediating sexual behavior. It acts at projections to septal nuclei from the ventral tegmental area, the bed nuclei, and the nucleus accumbens.²⁰ An occasional manifestation of dopamine toxicity is hypersexuality,⁴ an effect that may be mediated by dysfunction of septal circuitry.

In summary, we describe two patients with markedly increased sexual behavior following VP shunt placement and damage to the septal region. To our knowledge, this is a previously unreported complication of the management of hydrocephalus and is one of only a few descriptions of behavior changes associated with septal injury in humans. Experimental and clinical observations suggest that the septal nuclei form one locus on an amygdaloid-hypothalamic-septal circuit that mediates sexual behavior.

This project was supported by the Department of Veterans Affairs, by the French Foundation for Alzheimer's Research, and by an Alzheimer's Disease Core Center Grant from the National Institute on Aging, Bethesda, Md.

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