The Clinical Features and Outcome of Stupor*

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Introduction

Stupor is an unusual but striking phenomenon, generally recognized but difficult to define in precise clinical terms. A large literature exists on the diverse conditions in which it may occur, but there is little information on differentiating one cause from another, or on the prevalence of these causes. This insufficiency extends to the phenomenology of stupor, where descriptive criteria are not always adequate for separating stupor from allied states. Such difficulties are reflected in a current text book by Merskey and Tonge (1965). The authors only recognize stupor if there is total akinesis, where it is assumed that the diagnosis is schizophrenia. Depressive stupor is not recognized as an entity.

Aetiological concepts have been obscured by a controversy, never completely resolved, as to the nature of the underlying illness in stupor. Bleuler, following Kraepelin, maintained that stupor was mostly of schizophrenic origin. Later, in 1921, Hoch introduced the concept of Benign Stupor, meaning that these cases recovered completely and were suffering from manic-depressive psychosis. His attempt to differentiate benign and malignant forms was not convincing, and was later proved unreliable by Rachlin (1937). Of nineteen case records described by Hoch as Benign Stupor, thirteen were available to Rachlin (1935) for a followup study, and these patients had developed unequivocal evidence of schizophrenia. Conclusions based on these studies are limited by the small number of cases either described or later traced. and also by selective factors. For instance, cases located by Rachlin tended to be those with a

record of institutional care, thus favouring schizophrenia at the expense of depression.

An important contribution to this debate was Stanley Smith's (1959) survey of twenty-seven patients with stupor in an English mental hospital, in which he found that depression was commoner than schizophrenia. This was the first quantitative review of prevalence, but no cases with organic pathology were included and there was no follow-up. That this might be crucial is illustrated in a case report published by Hoenig and Toakley (1959). A patient with recurrent stupor presented as a mixed neurotic illness, and it was only at autopsy that a craniopharyngioma pressing on the brain stem was discovered. This suggests a neurophysiological basis for stupor through disturbance of the alerting mechanism of the reticulo-activating system. Cravioto's (1960) description of seven fatal cases with basilar artery thrombosis associated with infarcts of the brain stem lends further support to this type of theory.

It is clinical practice to scrutinize carefully any patient with stupor for a possible organic condition. Even so this may not be suspected. A fatal case, examined by the author, was attributable to hypoglycaemia from acute pancreatitis, as revealed (surprisingly) at autopsy. It has been long-established that stupor is not exclusive to the functional psychoses and that clinical recognition is difficult (Herman, 1942). There have been numerous references to the wide range of organic disorders in which stupor can occur, reviewed recently by Hoenig (1959).

Apparent hysterical stupor has also been described by Garmany (1955) and Neustatter (1942).

The present study was designed to overcome the objections to previous work in this field, which has been based on clinical impressions

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or, if quantifiable, on an insufficient number of cases. The scale of the project has been enlarged by investigating the outcome of 100 cases of stupor admitted to the Bethlem Royal and Maudsley Hospitals. The sample includes cases with organic pathology and offers a wider source of material than that available to previous workers, because admissions to the combined hospitals are enriched by referrals from other institutions.

It was hoped to test the validity of the underlying diagnosis by examining the outcome, and that an analysis of the characteristics of the sample, together with data on the modern course of the illness, would serve as a pattern for reference.

METHOD

Selection of Sample

Material was derived from admissions to the combined hospitals who exhibited stupor while under observation by medical staff. In the fourteen year period, 1948-61, some 250 individual patients were in stupor, comprising 1.6 per cent. of the total in-patient population. From these an arbitrary 100 were selected, composed of all the patients from one unit, supplemented by others chosen in a random manner.

Follow-up Technique

To every patient or close relative there was sent a standard letter explaining the purpose of the survey, and inviting co-operation by attendance at the hospital for interview, or, alternatively, a home visit by the author was offered. Replies were received from about 70 per cent., and the remainder were traced by approaching another informant, or by discovering a change of address from general practitioner records or from other hospitals dealing with re-admissions of many of the patients. A personal visit to the last known address was occasionally required.

As a result of thorough enquiries, 97 patients were traced whose outcome after discharge was known for a minimum follow-up period of two years, if they had survived. The small number of untraceable patients (3) and the fact that no more than one occurs in a diagnostic category made it unlikely that the operation of selective factors would bias the results based on the patients successfully traced.

Personal interview of both patient and informant was regarded as the most valid method of follow-up assessment, and once the patient had been traced this was achieved by a domiciliary visit, if necessary. Interview was arranged with 56 per cent. of patients, and in 44 per cent. this

included an informant. From the remainder who were traced, follow-up data were obtained by other methods:

- (i) Perusal of hospital records and correspondence, dealing with admissions subsequent to the key admission.
- (ii) Annual questionnaire is sent by post for 10 years to all patients admitted to the unit under survey. There is no experimental evidence of its reliability or validity, so that, mindful of this reservation, it was fortunate that for only four patients was this instrument the exclusive source of data. For the others, supplementary information was derived from
 - (iii) communication with general practitioner, and
- (iv) correspondence or telephone conversation with patient or informant.

The interview location was the patient's home in 19 cases, the hospital of residence in 12, and a place of mutual convenience, usually the Maudsley Hospital, in 25.

Failure to interview the traced survivors was largely due to geographic or occupational difficulties (11 patients); with 6 the present address remained unknown, and 5 specifically declined to co-operate, manifesting hostile or paranoid attitudes.

The design of the follow-up interview was mostly unstructured, but direct enquiry was sometimes necessary to clarify either the previous or current mental state and other points.

Physical examination was performed if indicated.

Selected Criteria of Stupor

In the present state of knowledge, stupor is defined in descriptive terms, the basic triad consisting of akinesis, mutism, and relative preservation of consciousness. In neurological parlance, akinetic mutism (Cairns et al., 1941) is equivalent to stupor. Movement of eyes and respiratory musculature is preserved. Often the absence of speech and movement is temporarily in abeyance, being restored as a result of external stimulation or for reasons that are obscure to the observer. This does not contradict the presence of stupor; it is an index of the degree of stupor. Thus an ideal definition would specify the means whereby speech and movement were temporarily restored; the extent to which they were restored; and for how long. But in practice these clinical distinctions are arbitrary, the state of stupor merging on the one hand with coma, and on the other with severe retardation.

The assumption that consciousness is relatively preserved is based on the presence of eye movements which appear purposeful rather than random, and the subsequent recall of events in the stuporose state which often occurs. However, spontaneous conjugate eye movements are found in mid-brain lesions, when they bear no relation to the level of consciousness, which may be totally impaired. Again, stupor may result in total amnesia, so that the factor of consciousness is difficult to assess in some instances. The disinhibitory effect of intravenous sodium amytal is variable in degree, and effective in severe retardation (and possibly in coma as well, were it to be tried).

Finally, it is salutary to consider what is clearly not stupor. We must exclude states of sleepiness (which imply potential arousal), loss of consciousness, and organic motor paralysis. Much depends on stimulus response as it affects the interpretation of the level of consciousness. A neurological opinion (Williams et al., 1951) is that the response to extreme painful stimulation is diagnostic. In stupor this elicits (i) blinking of seemingly alert eyes; (ii) a purposive reaction (pushing away the doctor's hand

holding the pin). A comatose patient would rub or scratch the tormented area, or make ineffectual protesting movements. Their conclusion that "stupor has a clear distinction from states of impaired consciousness such as confusion or coma" is untenable in much of psychiatric practice. It is sometimes held that because movement is suspended at such a high level of central nervous system activity the limbs will retain their normal tone of relaxation, but this does not take into account the rigidity of catatonic stupor.

Since these difficulties make it imperative to apply a reliable operational description, an attempt has been made to measure the severity of stupor and to differentiate it from other states. The various features have been scored as shown in Table I, data being chosen that could easily be gathered from the clinical records.

Thus, a patient in coma was excluded from the sample if all these data were identified: duration more than one day, sphincter disturbance, total akinesis and mutism, tube-

TABLE I

Description	Score	Coma	Total stupor	Partial stupor	Sleep
(i) Duration more than one day	I	I	ı or o	ı or o	ı or o
(ii) Sphincter disturbance (bladder or bowel)	I	I	ı or o	ı or o	0
(iii) Totally mute (iv) Partially mute (occasional monosyllabic	2	2	2 or 0	2 or 0	O
response to questions)	1	o	ı or o	ı or o	I
 (v) Totally akinetic (usually bedfast) (vi) Partially akinetic (fidgeting movements; or can be induced to move with verbal commands; or can be induced to co-operate 	2	2	2 or 0	2 or 0	o
in feeding, etc.)	I	0	ı or o	ı or o	1
(vii) Tube-feeding or intravenous nutrition (viii) No purposive response to nocuous stimula-	1	I	ı or o	ı or o	O
tion (ix) Stimulus response which is purposeful but	2	2	2 or 0	o	0
short of arousal	I	o	ı or o	I	o
(x) Eyes closed in spite of arousal stimuli	2	2	0	0	0
Maximum Score	12	11	10	8	3

The maximum score is 12, not 14, because the presence of total mutism excludes partial mutism; similarly, total akinesis excludes partial akinesis.

Coma score is 11 Total stupor score is 9 or 10 Partial stupor score is 8 to 4 Sleep states score, 3 or 2 feeding, non-purposive response to nocuous stimulation, and eyes remaining closed in spite of arousal stimuli.

A patient in total stupor will differ in respect to the appearance of the eyes, which are open and may be induced to follow a visual stimulus. Response to ordinary (non-painful) stimulation may be purposeful. To nocuous stimulation there could either be a non-purposive response, as in coma, or the reverse. Marginal latitude in the total stupor range of scores was permitted to the extent of dropping one point in the other items. In other words, the patient might be partially, rather than totally, akinetic.

Scores for partial stupor were given a larger range. Minimal features for inclusion in the sample might be partial akinesis and mutism; duration more than a day, no sphincter disturbance (patient can be induced to use toilet or bedpan); simple feeding by coaxing; and clearly open eyes with purposeful response to all stimulation.

Sleep has been included for completeness. A patient would have been excluded from the sample if stimulation elicited arousal. Sometimes this required considerable persistence, as in a patient with the Klein-Levin syndrome.

In selecting the stupor sample, it should be emphasized that occasionally an arbitrary element in the decision was unavoidable, however carefully the descriptive scoring criteria were applied to distinguish between sleep and stupor, coma and stupor, and pronounced retardation and stupor.

Diagnostic Categories

The following criteria were employed to simplify classification:

- (i) Depression. A predominantly affective disorder, excluding mixed neurosis (vide infra) and brain damage
- (ii) Mixed Neurotic. No cases of stupor in a purely hysterical setting were convincingly detected, but in order to segregate cases with a combination of manifest superficial motives in an assumed stress situation, plus hysterical features in the form of dissociation phenomena or exaggerated histrionic behaviour, the term "mixed neurotic" was introduced. These patients were, in addition, depressed to a greater or lesser degree. Alternatively, when hysterical features were superimposed on brain damage, the category of organic was applied. (iii) Organic is a self-explanatory title, embracing toxic-confusional states and irreversible

brain damage. The clinical diagnosis was often confirmed by special investigations, and in a high proportion of cases the cause of death was confirmed by autopsy.

- (iv) Schizophrenia. It is to be noted that acute schizophrenic reactions and cases designated as schizo-affective illness were included.
- (v) Uncertain. Here there were either unresolved diagnostic difficulties as a result of atypical features, or there was insufficient reliable information on which to base a firm diagnosis.

RESULTS

A. Demographic Characteristics of the Sample

TABLE II
Sex and Age

I	Age		Males	Females	Total
Under 30			19	24	43
30-39			19 6	6	12
40-49			6	9	15
50+	• •	• •	9	21	30
Tota	ıl	••	40	60	100

1. Sex and Age Distribution

The sample consists of a majority of females (60 per cent.) particularly in the over 50 age group (70 per cent.), while for both sexes the largest distribution is in the under 30 age group. When compared with the total hospital inpatient population for the same period, there is no statistically significant difference.

Stanley Smith's small series revealed an impressive majority of females (81 per cent.), notably in his depressive group (92 per cent.) and with schizophrenics (89 per cent.). These figures cannot be directly compared with the present sample, which is influenced by selective factors in the admission policy. However, sex distribution in relation to the diagnosis, finally revised at follow-up if possible, was examined in the present sample (Table III).

TABLE III

Final Diagnosi	s	Male	Female
Depression Mixed Neurotic Organic Schizophrenia Uncertain	••	 9 3 9	16 7 14 19

Again a female majority was found, chiefly 64 per cent. in depression and 70 per cent. in mixed neurotic stupor. When the female risk for depressive stupor is compared with the hospital population for psychotic and neurotic depression, taken together, there is no significant difference.

2. Occupation

This was not regarded as significant, in that the sample bore a close resemblance to the hospital in-patient population in general.

3. Marital State at Key Admission

Similar observations apply to the distribution by marital state. There was a high incidence of single persons in the sample (36 per cent.), attributable to the high proportion of schizophrenic patients, only a minority of whom were married.

B. Diagnostic Formulation at Key Admission

Table IV describes the incidence of the diagnostic groups in the 100 patient sample.

Table IV

Formulation at Key Admission (100 cases), by Psychiatrist
at In-patient Discharge

I	Diagn	osis		No. cases
Depression			 	25
Mixed Neuroti	c		 	10
Organic			 	20
Schizophrenia			 	31
Uncertain			 	14

The diagnosis of organic pathology was substantiated in 20 per cent. of the cases. This high proportion, compared with the overall number of cases admitted to the combined hospitals with organic states, represents selective factors favouring the admission of these interesting patients. Two examples illustrate this (Cases 1 and 2 in the Appendix).

The tendency for patients with organic stupor to be deflected from general hospitals can be further understood in the light of the presenting clinical picture, often a confusional state, but sometimes an affective disorder.

The nature of the organic disturbance varied, as shown in Table V, where the diagnosis is that recorded by the psychiatrist at the time of in-patient discharge.

Of the functional psychoses, the largest group were the schizophrenic patients, 31 per cent. of the sample, followed by those with depression, 25 per cent.

Mixed neurosis, which involved hysterical features, accounted for 10 per cent. of the

Table V

Key Admission I		No. cases		
Intracranial tumour	• • •	•••	•••	2
Pre-senile dementia				4
Arteriopathic dement	ia			ī
Senile dementia				2
Post-encephalitic state	e			2
Confusional state				4
Neurosyphilis				3
Cerebral cyst				ī
Tuberose sclerosis (po	st-epi	leptic st	ate)	I
Total	•••		••	20

sample. A typical case was an immature Swiss peasant girl, admitted to hospital after a suicidal attempt. For several weeks she was mute, lying and falling on the floor, overbreathing and displaying bizarre fits. There was fluctuating stupor, but at times she could be coaxed out of bed, screaming noisily. Hysterical features existed in a setting of depression.

Finally the group classified as uncertain, numbering 14 per cent., was unusually large in comparison with hospital admissions in general. These patients often presented great diagnostic difficulty, sometimes clarified by follow-up assessment. (See cases 3 and 4 in appendix.)

In some cases the cause of the diagnostic uncertainty was the distinction between a toxic-confusional episode (or post-epileptic state) and a functional psychosis. In these unresolved situations the results of special investigations were inconclusive. As the spectrum of investigations, even with access to neurosurgical facilities, is relatively crude, often ambiguous in interpretation, and likely only to detect gross lesions, it is interesting that only one case recorded as uncertain was later assessed as organic after follow-up, and then by virtue of psychiatric assessment.

It might be supposed that the very nature of stupor would constitute a diagnostic obstacle, because the patient could make no verbal contribution to the diagnosis, and recollection after the event could be falsified, if not totally lacking. In actual practice, these considerations did not seem to be a handicap for the following reasons: reliable information from persons

other than the patient; release of speech after disinhibition with sodium amytal, the form and context of which might be diagnostically useful; and the fact that under modern methods of treatment stupor was rarely prolonged and usually left in its wake characteristic features of the underlying illness. The recall of stupor sometimes revealed delusional motivation of diagnostic value.

C. Special Physical Investigations

To test the significance of the results of physical investigations in terms of the outcome of the illness, assuming an organic lesion, if progressive, would declare itself in time, the number of cases were counted in which investigations produced abnormal findings. They totalled 31. Investigations included EEG, air studies, biochemical measurements, etc. Since there were originally twenty organic cases, this suggests that in eleven cases there was a possibility of re-classifying functional illness at follow-up. However, the excess of patients with abnormal findings by investigation over those with known or assumed organic cerebral disturbance depends on non-specific EEG abnormalities, common in schizophrenic catatonic stupor. In fact, where re-classification to organic was justified by longitudinal assessment, investigations were unhelpful; clinical examination was decisive.

D. Previous Personality

Work by Weinstein et al. (1955) on 23 patients with brain damage suggested that the reaction of withdrawal, muteness, and inattention was characteristic of a particular personality type where a regressive reaction (so-called) under stress was a basic pattern of behaviour and conduct. This is a tempting hypothesis, but one difficult to substantiate without a reliable psychological instrument to measure personality. Weinstein investigated this aspect of behaviour in the pre-morbid personality by talking to relatives, noting the replies to such questions as "How do you react when upset?" Significant answers were: "She'd give you the silent treatment." "He would never argue; just clam up." "She'd get quiet and then you would know she was angry." This form of clinical impression was not applicable to the present series, where only half the total patients were subject to interview. Therefore as an approximation to this postulated personality factor of withdrawal, search was made for descriptions such as "autistic", "withdrawn" and "solitary", if concrete examples of the habitual use of withdrawal and avoidance in stressful situations were not present in the records. On this crude basis a total of 43 of the patients in this study showed pre-morbid withdrawal features, shown in Table VI, column 1, by diagnostic grouping.

Since the shut-in seclusive type of personality (described by Bleuler as schizoid) so common in

TABLE VI

:	Diagn	osis			1. Previous Personality of Withdrawal	2. Total Stupor	3. Mortality in Key Illness	4. Multiple Attacks of Stupor
Depression				•••	6	6	3	6
Mixed Neurotic					I	0	0	3
Organic					5	5	7	2
Schizophrenic					28	6	I	ΙΙ
Uncertain	• •	• •	• •	• •	3	I	I	O
Total					43	18	12	22

In Column 4, alone, the diagnosis is the final one, revised at follow-up if necessary. Otherwise, the diagnosis is that recorded by the psychiatrist at time of in-patient discharge.

schizophrenia may be indistinguishable from the personality that withdraws in reaction to stress, it is not surprising that 28 of the 33 are schizophrenics. It could be argued, "Why are not stupor reactions commoner in view of the incidence at large of schizophrenics with previous schizoid personality?" Or "Why are stupor reactions almost equally common in depressives as in schizophrenics, if the withdrawn personality is unusual in depression?" The answers to this issue lie in more precise personality measurement by specially constructed and validated questionnaires.

If this concept is valid, and stupor is an unusual stress reaction in a predisposed personality, then there should be a high incidence of stress as a precipitant of the condition. When such factors were thought to be operating they were recorded in the original formulations and could be confirmed at follow-up. Attention was paid to:

- (a) correspondence in time between onset of symptoms and significant environmental stress;
- (b) termination of stupor, as a probable result of psychological events, when this occurred (3 per cent. of cases);
- (c) remission of illness associated with environmental change, i.e. dramatic improvement only after discharge from hospital.

In fact, psychological precipitating factors were only elicited in 15 cases.

E. The Characteristics of Stupor

1. Severity

After applying the system of scoring, total stupor was identified in 18 per cent, and partial stupor in 82 per cent., of the sample. The division of total stupor into diagnostic groupings is shown in Table VI, column 2.

The five organic cases consisted of pre-senile dementia, tumour on the dominant frontal lobe, cerebrovascular disease, diffuse brain damage resulting from prolonged hypoglycaemia, and symptomatic psychosis of uncertain origin.

Lewis (1934), in his comprehensive clinical

survey of depression, noted that severe stupor, as indicated by total mutism, tube-feeding, and possible incontinence, was a rare event. The findings of this survey indicate that severe catatonic stupor is a comparably rare manifestation of schizophrenia. But severe stupor is not more common in catatonic than in depressive stupor.

None of the patients with a diagnosis of mixed neurosis fulfilled the criteria for total stupor. It seemed that there was marked fluctuation in the depth of stupor over a few hours, severe stupor, if present, was never prolonged.

2. Duration

The duration of stupor is shown in Table VII, the number of cases gradually diminishing with time, the largest number terminating within a week. This contrasts with the protracted course of stupor of twenty and more years ago, and reflects the active therapeutic approach of modern psychiatry.

TABLE VII
Stupor Duration

Duration	0-1 wk.	ı wk- 4 wks	1 mth- 6 mths	6 mths+
No cases	43	43	16	4

The four patients in stupor for six months or more suffered from brain damage, e.g. cerebral atrophy resulting from prolonged hypoglycaemia, cerebral atrophy presumed to be arteriopathic, frontal lobe tumour, and postencephalitic state. In this latter condition, stupor may occur in 1 per cent. of the cases; the patient in this series has been in stupor for seven years, interspersed with periodic alertness.

3. Means of Termination of Stupor

These are classified in Table VIII.

In approximately one-third of the cases of stupor there is a spontaneous cessation. Physical treatment was associated with termination in 53 per cent. of the cases. The quickest and most effective method was electroplexy, which was, in fact, the most commonly employed. Some of

Table	VIII
Termination	of Stupor

	Termi	nation		 Spontaneous		Psychological Factors	Death
Number of Cases			 	 36	53	3	8

the cases receiving treatment over ten years ago responded to a course of insulin coma. Other patients, in the last decade, have benefited from the development of the phenothiazine group of drugs, and such therapy was the exclusive means of terminating stupor in a few cases.

In one patient, the intravenous use of sodium amytal appeared to result in a sustained improvement of the stupor state. However, the usual response to this technique of disinhibition was to release only a minor degree of speech and movement, followed quickly by relapse. The content of this evoked reaction was either depressive, manifestly schizophrenic, or unspectacularly normal.

In 8 per cent. of cases there was a fatal outcome to the stupor attack. One schizophrenic girl of 15 died on the third day of catatonic stupor from respiratory infection. She had been receiving reserpine, known to impair the physiological reaction to stress by interfering with catechol amine metabolism. This is of debatable significance. Another patient was a youth of 20, dying in the third week of a fulminating illness, who presented the features of catatonic stupor with flexibilitas cerea for nine days before death from respiratory infection. There were difficulties in accepting this illness as functional, because of clinical features suggesting Cushing's disease. In fact, this case is recorded as "uncertain", since death occurred before investigations could substantiate a diagnosis of Cushing's psychosis. In the other six patients where death occurred in stupor, there was definite evidence of brain damage. The immediate cause of death was intercurrent infection or, in one case, myocardial infarction.

Lastly, 3 per cent. of the cases merited the conclusion that the termination of the state of stupor was linked with the operation of psychological factors. This is illustrated by the case of a 48-year-old married woman of neurotic

constitution and low intelligence, who developed a depressive illness which corresponded in time with depression and stupor in her newly-married son. The mother experienced recurrent episodes of stupor, never lasting more than three hours, in which there was akinetic mutism, with incontinence of urine. Physical treatment was not given, the attacks ceasing pari passu with relief of depression, as soon as the son's condition improved.

F. Duration of Key Illness

Table IX shows the number of cases corresponding to set periods of illness. In only one patient was the illness lifelong; he suffered from classical tuberose sclerosis, with persisting neurological signs and epilepsy. The majority of patients experienced full remission within two years, from which it can be concluded that the tendency towards chronicity of illness with stupor is no greater than with an unselected series of admissions to the hospital.

TABLE IX

Duration of Illness: Total Period before Death or Recovery

Period				No. cases
Under 6 months				36
6 months to 2 years	• •	• •	• •	3 6
2 years to 5 years	• •		• •	12
More than 5 years				15
Lifelong				I
Total	• •	• •	••	100

A difficult problem in measuring the duration of any mental illness is the assessment of degree of recovery. This almost certainly requires personal examination to distinguish a complete recovery from one which may be complete for social and practical purposes but falls short of complete restitution.

G. Mortality in Key Illnesss

Table VI, column 3 indicates the serious implications of the state of stupor, with reference to underlying diagnosis. Eight of the 12 deaths occurred during stupor, and have been mentioned previously. Three patients suffering from depressive illness successfully committed suicide, nine days, six months and seven months, respectively, after discharge from hospital.

H. Progress after Discharge

1. Period of Surveillance

Patients qualified for inclusion in the sample if their progress could be followed for at least two years. A period up to five years accounted for 38 per cent. of patients, and 37 per cent. were surveyed for six to ten years. The remainder were followed for eleven or more years.

2. Multiple Attacks of Stupor

A total of 22 patients suffered multiple attacks of stupor, either before, during, or after the key admission. Table VI, column 4, records this in terms of the diagnostic groups.

Multiple attacks were judged on the basis of:

- (a) circumscribed recurrence of the illness with stupor; or
- (b) significant remission of the mental state between attacks of stupor (as in an 18year-old girl with five periods of depressive stupor, occurring at mid-cycle, with full remission between attacks).

It will be observed that patients with schizophrenia are almost twice as numerous as those with depression, which is partly accounted for by the larger number of schizophrenics in the sample. As 32 patients were finally classified as schizophrenic, the likelihood of multiple attacks in this illness is roughly one in three. Similarly for depressive illness, the likelihood is one in three and a half.

Multiple attacks of stupor were usually two in number, for both schizophrenics and depressives. The largest number of attacks was six, suffered by a patient with schizophrenia.

Regarding the interval between the first and second attacks of stupor, this was between one and six months in two patients (depression), but in the rest it was between one and four years.

It is of interest that no cases in the uncertain group suffered previous stupor episodes, implying that this is of diagnostic assistance.

3. Recall of Stupor

Table X attempts to describe the degree of recall of thought content and external events while in the state of stupor. Memory data were not always recorded in the notes of the key illness; hence the largest group of 41 per cent. comprises the patients in whom recall is unrecorded, or unknown from the follow-up material available.

In the rest, memory of the stupor event was at least two years old and was assessed in relation to memory of other events of the key illness. This was supplemented by data on immediate memory, when available in the records. Thirty-one per cent. of patients had total amnesia for the stupor period. Hence, the inference that consciousness is relatively preserved in stupor, which is based on the recall of events, is not reliable in itself. Partial recollection of events occurred in 10 per cent., while full recall was restricted to 18 per cent. of the patients.

What is experienced in the state of stupor? Jaspers writes: "These psychic states are puzzling. We do not know how these patients feel. We have hardly any self-description. If they describe themselves, they use words reminiscent of meaningful states in our own existence but

TABLE X

Recall of Stupor either at Key Admission or on Follow-up

	Rec	all			Total, i.e. no amnesia	Partial amnesia	Nil, i.e. total amnesia	Unrecorded or unknown
Percentage			••	 	18	10	31	41

probably they can only be taken as analogies."

When patients in this survey were able to verbalize their feelings, it was usually in terms of fear of an alien environment. A few illustrative quotations are:

"Afraid of everyone."

"Behaving like a baby because I couldn't adjust to a strange environment."

"No interest; I didn't care."

"I was just frightened to ask where to go [micturition]. I wanted to rest—to be left alone—not to be bothered."

"I didn't want to be in hospital."

These remarks confirm Jaspers' view; they seem to be poorly elaborated echoes of the original experience. It was unusual to establish the fact that the patient experienced akinesis and mutism as a volitional act, with delusional motivation.

4. Outcome Status

Table XI shows the outcome status of 86 traceable patients who survived for a two-year follow-up assessment, together with eleven patients who died within two years of stupor, to underline the heavy mortality associated with this type of illness (19 per cent. of original sample).

Outcome is considered under four headings: (a) Dead. Mortality in the key illness has been previously described in twelve patients. In one, suffering from pre-senile dementia, death occurred more than two years after admission for stupor.

Seven patients died after the key illness had finished. Again the organic group predominates. One 68-year-old female died as a result of diabetic gangrene. The original stupor had been in the setting of a toxic-confusional state, associated with a cataract operation and uncontrolled diabetes. Another, a 28-year-old male, died from the effects of a cerebral cyst.

In two cases, it was considered that death was unrelated to the cause of the stupor, i.e. a 73-year-old male dying six years after depressive stupor of ischaemic heart disease, and a 61-year-old female dying three years after a presumed mixed neurotic stupor of hepatic carcinoma, confirmed at autopsy. But the issue was not always clear (Case 5 in Appendix).

For the sample as a whole there were four fatal suicides, three from depression and one from schizophrenia.

(b) Resident in hospital or institution, due to nature of mental illness and/or severity of social disturbance. This accounted for about one-sixth of the 86 two year survivors, with organic cases preponderant.

Mixed neurotic cases were absent from this institutionalized group. There was no significant difference between schizophrenia and depression.

TABLE XI

Outcome in Relation to Diagnosis, revised if necessary

Outcome Status	Depression	Schizo- phrenia	Mixed Neurotic	Uncertain	Organic	Total
Resident in hospital or institution	4	3	0	I	6	14
Returned to pre-morbid personality level	12	12	4	I	4	33
Handicapped, i.e. personality change, social disturbance	4	16	4	4	3	31
Dead	4	2	I	3	9	19
Total	24	33	9	9	22	97

(c) Handicapped: i.e. personality change; social disturbance. The personality damage being of insufficient degree to warrant custodial care, the patients were adjusted in an appropriate community environment, often in sheltered employment. A few patients in this group were considered to be mentally ill, but eschewed psychiatric help, being afraid of committal to hospital. The striking result is that half of the schizophrenic two year survivors were so handicapped, in contrast to under one-fifth of those with depression.

Thus, depressive and schizophrenic stupor fulfilled the prognostic implications of the underlying illness, the difference being significant at the 2 per cent. level.

Forty-four per cent. of the mixed neurotic patients were handicapped at follow-up assessment, compared with 19 per cent. for depression, but this difference is not statistically significant.

(d) Restitution to pre-morbid personality. Assessment of full remission without personality change accounted for the largest outcome group (33 patients).

Patients with depression number 12, and schizophrenia 12; comprising 57 per cent. and 38 per cent. respectively of those patients so diagnosed in the follow-up sample. This difference is not significant.

Interpretation of all these results in the light of the prognostic significance of stupor is difficult, without data on the symptomatic outcome of comparable patients without stupor. A further difficulty is the current trend towards community care of chronic patients who can be rehabilitated, which may be reflected in the small number of institutionalized schizophrenics in the sample.

All the characteristics of the material so far described were related to outcome status. The following findings were observed:

(i) Severity of stupor. Five patients with total stupor had died within two years (4 brain damage: 1 schizophrenia). A sixth, presenting with a confusional state associated with diabetes mellitus, died four years later of diabetic gangrene. Of the 12 patients surviving, total stupor, 8 were fully recovered (67 per cent.), compared with 25 out of 66 in the case of partial

stupor (38 per cent.). The difference is not significant $(P=\cdot 06)$ but does suggest an interesting trend: the deep stupors recover best.

Severity of stupor related to the other outcome groups revealed less marked differences.

- (ii) Duration of stupor. For periods longer than four weeks, the numbers were too small to yield significant results. But selecting two periods, one less than a week and one greater, there was no statistically significant difference in outcome.
- (iii) Sex and age figures, when related to outcome status, showed no statistically significant difference.
- (iv) Multiple attacks of stupor, when compared with solitary attacks, did not result in a different outcome.

I. Diagnostic Formulation at Time of Follow-up

Table XII expresses the revised diagnosis, assessed at follow-up by longitudinal study, in relation to the original diagnosis at key admission. Total number of patients considered is 86. Excluded are the 3 untraceable, and the 11 patients who did not survive for a two-year follow-up, in none of whom was the diagnosis found to be altered.

In 13 cases there was no correlation between original and revised diagnosis. These are briefly reviewed.

1st. "Uncertain" revised to a definite diagnosis; depression in 2, schizophrenia in 4, and organic in one of the cases. However, 6 of the "uncertain" continued to be diagnostically unresolved and thus remained "uncertain".

Where the diagnosis was revised to depression, the mental state in the key illness had led to uncertainty because of the presence of clouding of consciousness and incongruity of affect.

In the patients revised to schizophrenia, the clinical picture in the key illness had been atypical; in two, affective features were prominent; in a third, confusion due to drug addiction complicated the mental state; in a fourth, signs of Parkinsonism suggested organic psychosis.

As regards the patient reclassified to organic, this was a 56-year-old woman, with total stupor of one month's duration, refractory to E.C.T.

Table	XII
Changes of	Diagnosis

On Admission		On follow-up				
	Depression	Mixed neurosis	Organic psychosis	Schizo- phrenia	Uncertain	Change of diagnosis
Depression 2	18		(2)	(1)		+3, -3
Mixed Neurosis Organic Psychosis	9 1	9	13		(1)	+3, -1
Schizophrenia 2 Uncertain 1	· ; ;		(1)	27 (4)	(1) 6	+3, -1 $+5, -2$ $+2, -7$
Total 8		9	16	32	8	86

Numbers in brackets represent changes in diagnosis

Personality deterioration suggested pre-senile dementia, but full investigation, including cortical biopsy, was inconclusive. However, follow-up assessment confirmed this diagnosis.

2nd. Schizophrenia revised to "uncertain" in one patient because of inadequate follow-up information. Assessment was by questionnaire and general practitioner records, but interview was not possible, though considered crucial in this case.

Schizophrenia revised to depressive illness in one patient, on the grounds of sustained restitution of personality after a key illness of four years' duration. Affective features were prominent, suggesting mixed affective psychosis in retrospect.

3rd. Depression revised to schizophrenia: the original presentation was exclusively affective, but subsequently affective incongruity and thinking disorder developed, characteristic of schizophrenia.

Although depression was changed to organic in two cases (Cases 6 and 7 in Appendix), it seems that the original illness might have been different from the later dementing illness.

4th. Organic revised to uncertain: a patient developing stupor in a toxic-confusional state; follow-up data were inadequate without interview.

Discussion

The contribution of a follow-up study to the diagnosis of stupor has revealed 13 patients in the sample in whose cases retrospective and

current examination were discordant. This figure does not challenge the general validity of the original diagnosis, bearing in mind the problems of psychiatric assessment and the lack of agreement among trained observers, as described in the U.S.A. by Schmidt and Fonda (1956) and in this country by Kreitman et al. (1961).

In only one patient was a diagnosis of depression revised to one of schizophrenia. This stands in contrast with Rachlin's finding that in a high proportion of Hoch's cases originally diagnosed as Benign Stupor, the eventual state on follow-up showed itself as schizophrenic. The present author cannot agree that general conclusions can be based on Rachlin's findings. Depressive stupor is a valid entity; it is nearly as common as schizophrenic stupor and is just as likely to be profound. Only one patient presented a clinical picture of benign stupor comparable with the description in Hoch's series. Here, treatment had been withheld for research purposes. In fact, prolonged nonorganic stupor must be rare to occur under a modern active therapeutic regime.

Four patients classified as "uncertain" were revised at follow-up to a definite diagnosis of schizophrenia. In this series, the risk of underestimating the frequency of schizophrenic stupor might arise from employing stringent diagnostic criteria for this illness.

The literature on hysterical stupor is scanty. It has been described as a brief phenomenon in practised hysterics (Garmany, 1955), in pri-

soners, and in war neurosis. Neustatter (1942) fully documented a case of possible hysterical stupor of one month's duration in a solitary young soldier. This patient, however, had "Schnauzkrampf" as one of his symptoms; only a follow-up study could establish whether schizophrenia was the underlying illness. In the mixed neurotic group in the present sample, longitudinal assessment was in total agreement with the original diagnosis. Stupor, as an hysterical state, was one manifestation among others of disproportionate behaviour which could be attributed to dissociation. It was never isolated, however, being combined with either organic or depressive features.

No convincing cases were found at follow-up to indicate an organic lesion as an unknown cause of the stupor. Intra-cranial tumours in two patients were in different sites, so this shed no light on a possible neuro-anatomical mechanism in stupor. In the present series the features of the stupor state were identical, whether organically determined or not, and the diverse conditions associated with stupor cannot be easily integrated into a unifying theory.

Anatomical, physiological, chemical, and psychological theories have been advanced to explain stupor, as reviewed by Smith (1959). Multiple aetiology is consistent with Holland de Jong's (1956) conclusions, based on experimental catatonia in animals, that stupor is a non-specific, general reaction of the central nervous system. Presumably, some individuals are predisposed to develop this state. They might, to take two examples, be distinguished chemically by paradoxical reaction to adrenaline (Breggin, 1965), which could be experimentally tested, and/or by the recognition of a specific constitutional type. Crude analysis of the present sample in terms of pre-morbid personality does not suggest a specific personality trait predisposing to stupor, but perhaps more precise measurements are required. Once developed, there are no characteristics of stupor per se which reliably differentiate nosological groups. The diagnosis is made on the basis of other clinical features, and follow-up can be crucial in a minority.

The results suggest that the outcome in a

patient with stupor depends on the underlying illness. Organic causes are mostly irreversible, and therefore serious. Depressive and schizophrenic stupor fulfilled the prognostic implications of these disorders. There is a trend suggesting that the chance of full recovery is greater in total than in partial stupor, which invites the hypothesis that deep stupor may be a protective influence for those surviving.

However, the prognostic implications of stupor, as such, complicating an illness, could only be tested by matching similar cases where stupor was absent. Nevertheless, this survey provides a reference for broad predictions about an individual case of stupor that might arise in current practice.

SUMMARY

- 1. One hundred cases of stupor were surveyed retrospectively, and investigated by follow-up methods. Only 3 per cent. were untraceable.
- 2. The criteria of stupor are scrutinized, and a rating scale devised to distinguish total and partial stupor and isolate them from allied states.
- 3. Diagnostic incidence was: depression 25 per cent.; schizophrenia 31 per cent.; mixed neurotic 10 per cent.; organic 20 per cent.; uncertain 14 per cent.
- 4. Demographic characteristics of the sample are discussed.
- 5. Constitutional factors are assessed. The evidence for a specific personality trait predisposing to stupor is unimpressive.
- 6. Total stupor occurred in 18 per cent. of the sample. It bears no diagnostic significance in differentiating schizophrenia from depression, but there is a suggestion that it is associated with a more favourable outcome than partial stupor.
- 7. In the key illness, mortality was 11 per cent. Afterwards it was 8 per cent. Mortality was most prevalent in organic cases.
- 8. The likelihood of recurrent attacks of stupor with schizophrenia and depression is approximately one in three.
- 9. Total failure to recall external events, while in the state of stupor, occurred in 31 patients. The significance of this is discussed.

- 10. Symptomatic outcome at time of follow-up is described, and related to the various characteristics of the sample.
- 11. Retrospective diagnosis was compared with follow-up assessment. Discordance in 13 cases was analysed. In only one case was depression re-classified as schizophrenia, so that the concept of Benign Stupor in depressive psychosis remains valid, and this was confirmed by a better prognosis compared with schizophrenia.
- 12. Stupor as an hysterical phenomenon was not encountered in isolation, being combined with either depressive or organic factors.

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REFERENCES

BLEULER, E. (1949). Lehrbuch der Psychiatrie, 8th ed. p. 387. Berlin: Springer-Verlag.

Breggin, P. (1965). "Sedative-like effect of epinephrine." Arch. gen. Psychiat. (Chic.), 12, 255-259.

CAIRNS, H. et al. (1941). Brain, 64, 273.

CRAVIOTO, H., SILBERMAN, J., and FEIGEN, I. (1960). "A clinical and pathological study of akinetic mutism." Neurology (Minneap.), 10, 10-21.

GARMANY, G. (1955). "Acute anxiety and hysteria." Brit. med. J., ii, 115.

HERMAN, M., HARPHAM, D., and ROSENBLUM, M. (1942). "Non-schizophrenic catatonic states." N. Y. St. J. Med., 42, 624-627.

HOLLAND DE JONG, H. (1956). "Experimental catatonia in animals and induced catatonic stupor in man." Dis. nerv. Syst. (April) 17, 135-139.

HOCH, A. (1921). Benign Stupors: a Study of a New Manicdepressive Reaction Type. Cambridge Univ. Press.

HOENIG, J., and TOAKLEY, J. G. (1959). "The diagnosis of stupor." Psychiat. et Neurol., 137, 128-144.

JASPERS, K. (1913-1946). Allgemeine Psychopathologie. Translated as General Psychopathology 1962. Pp. 601-602.

KREITMAN, N., SAINSBURY, P., MORRISSEY, J., TOWERS, J., and Scrivener, J. (1961). "The reliability of psychiatric assessment: an analysis." J. ment. Sci., 107, 887-908.

Lewis, A. J. (1934). "Melancholia: a clinical survey of depressive states." J. ment. Sci., 80, 277-378.

Merskey, H., and Lawton Tonge, W. (1965). Psychiatric Illness. 226–227. London.

NEUSTATTER, W. L. (1942). "A case of hysterical stupor recovering after cardiazol treatment." J. ment. Sci., 88, 440-443.

RACHLIN, H. L. (1935). "A follow-up study of Hoch's 'benign stupor' cases." Amer. J. Psychiat., 92, 531-558. - (1937). "A statistical study of benign stupor in five

State hospitals." Psychiat. Quart., 11, 436-444.
SCHMIDT, H. O., and FONDA, C. P. (1956). "The reliability of psychiatric diagnosis: a new look." J. abnorm. soc. Psychol., 52, 262-267.

SMITH, S. (1959). "An investigation and survey of 27 cases of akinesis with mutism (stupor)." 7. ment. Sci., 105, 1088-1094.

Weinstein, E. A., Kahn, R. C., Slote, W. H. (1955). "Withdrawal, inattention, and pain asymbolia." Arch. Neurol. (Chic.). 74, 235-248.

WILLIAMS, D., and PARSONS-SMITH, G. (1951). "Thalamic activity in stupor." Brain, 74, 377-398.

APPENDIX

Case I

A 52-year-old woman with a severe frontal lobe disturbance due to a large dominant-hemisphere glioma. Apathy, present on admission, gradually progressed towards total stupor, continuous for seven months until she died. She was totally mute and inert, developing decubitus ulceration (a rare event in stupor). There was double incontinence. Liquids were swallowed if placed in the mouth. The preservation of consciousness was inferred from response to painful stimuli and the turning of the eyes in the direction of sound from a human voice.

Case II

A 57-year-old woman with a post-mortem diagnosis of cerebellar atrophy, postulated to be the sequel of encephalitis. Four months before death she exhibited periodic partial stupor. Although the mental state was similar to the previous case, there were times when she issued a torrent of abusive speech, and others when she was inaccessible, not responding to commands. It seemed reasonable to consider the predominant mental state as one of stupor, partial in degree, because both akinesis and mutism were not sustained beyond a few hours. In fact, she improved spontaneously, walking out of hospital, only to die one week after discharge.

A 56-year-old woman suffered three separate episodes of stupor over a period of two years while under hospital observation. Although depression was prominent, and confirmed by morbid thought content released by disinhibition with intravenous sodium amytal, there were some schizophrenic features, viz. inappropriate affect and rigidity (?catatonic). In one episode there was clouding of consciousness, an equivocal unilateral extensor plantar response, and an abnormal EEG focus which was absent on subsequent records.

In this patient there were unresolved diagnostic problems in the key illness, although the history of previous illnesses revealed two discrete depressive states. Follow-up assessment by personal interview, eight years later, disclosed that she had been recurrently depressed, and occasionally elated, over this period, with numerous attacks of stupor responding to E.C.T. Current examination showed an obese, jolly, warm-hearted woman, with no evidence of dementia or personality change (as stated by husband). There was amnesia for the stupor episodes. Manic-depressive psychosis seemed an indisputable diagnosis.

Case IV

A 41-year-old man from a cultured background, who had been suffering from narcolepsy for fifteen years. One month before admission he had exhibited attacks of mutism, rigidity, automatism, staring, grimacing, and upward rolling of the eyes. Parkinsonism of presumed post-encephalitic origin was strongly suspected, in view of mask-like facies, festination, bradykinesia, and blepharoclonus. However, catatonic schizophrenia appeared to be a plausible alternative to explain discontinuity of speech and recurrent attacks of transient stupor lasting approximately twenty minutes. In these he would be akinetic, either sitting stiffly in bed or standing still, mute and negativistic.

Follow-up seven years later showed that he had remained in a mental hospital as a chronic patient because of deterioration of social habits. At a personal interview, he showed mannerisms, was disconnected in talk, and inappropriate in affect. He had an excellent memory, was correctly orientated, and appeared above average in intelligence. There had been no stupor for seven years, and he explained the original episodes as deliberate attempts to be "awkward", alluding to a hostile environment. Current assessment was a chronic, deteriorated schizophrenic in whom the previous episode of Parkinsonism was an isolated one, and therefore unlikely to be postencephalitic. Drug-induced Parkinsonism is, however, worthy of consideration, although the symptoms were not dispelled by the use of chemical antidotes.

Case V

A 23-year-old female diabetic, in stupor for twelve days, finally terminated by intravenous sodium amytal. The diagnosis was uncertain, being possibly a schizo-affective illness, in view of depression, bewilderment, and bizarre delusions. She recovered rapidly and completely, only to die, two years later, suddenly from asphyxia due to inhalation of blood resulting from a tongue laceration in

an epileptiform fit. There was no previous history of convulsions or of hypoglycaemic states. Coroner's autopsy reported the brain as normal, and unfortunately no further information could be elicited.

Case VI

A 27-year old married man, insurance agent. Personality perfectionist: depressive phases. Key illness, one and a half years' duration. Depression with episode of total stupor. Good recovery. Progress: at age 49, re-admitted to hospital with nine days' depression and confusion: episode of partial stupor. Bizarre thought content, with abnormal mental state which never improved.

Follow-up 36 years after key illness, by interview with patient and wife. Continuous hospitalization previous ten years. Apathetic, unco-operative, attempts at suicide. Some deterioration of habits. Examination: spasticity with extensor plantar responses. Unkempt, retarded, apathetic. Marked disorientation in setting of clear consciousness. Gross failure of comprehension, with bizarre ideas. Revised diagnosis: pre-senile dementia. Key illness of depression presumably functional and quite separate from disease starting 22 years later, although this presented with depressive content.

Case VII

A 67-year-old widow at key illness. Domestic work. Personality: anxious, rigid and obstinate. Since age 21, five discrete episodes of confusional state following influenza, thrombo-phlebitis, and cholecystitis. At 58, short depressive reaction associated with suicide attempt. Key illness of three months' duration, nine years later. Depression with psychomotor retardation; partial stupor for one week; good response to E.C.T. Diagnosis: depressive illness. Progress: recurring episodes of depression and hypomania. No home of her own: resident in L.C.C. home. Follow-up examination: elderly woman aged 71. Apprehensive, suspicious, not depressed. Orientation incorrect. Impairment of memory for recent events, e.g. no recollection of Maudsley Hospital admission. Revised diagnosis: senile dementia. Possible that depressive stupor was associated with brain damage or a separate illness.

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