

Tautologous Diabetic Coma—A Behavior Syndrome

Multiple Unnecessary Episodes of Diabetic Coma

Franklin B. Peck, Sr., M.D., and Franklin B. Peck, Jr., M.D.,† Indianapolis*

The occurrence in a single patient of almost 50 repetitive instances of severe diabetic coma within 8 years of onset of the disease at age 12 is unique. Multiple comas of reasonable number are not unusual, but we know of no reported case having so many. The term tautologous has not been previously applied to diabetic coma. It is usually employed rhetorically, but in this instance it seems especially apropos, signifying as it does "needless repetition." The development of this behavior pattern is of special interest in relation to the effect of stressful situations on the control of diabetes and the influence of subsequent poor control on the development of vascular complications.

CASE HISTORY

The patient, (M.R.J.) 70896, a white female, aged 20, was first admitted to Indianapolis General Hospital in 1946 when she was 12 years old. She was unconscious, in deep diabetic keto-acidosis following three months of prodromal symptoms of hunger, thirst, and polyuria, with loss of 10 pounds in weight. Blood sugar was 500 mg. per cent, plasma carbon dioxide combining power 5 mEq./L., and Rabinowitch severity index 20¹ (very severe). In the ensuing 12 hours 500 units of insulin were required to restore normal metabolic equilibrium. During the succeeding days good balance was established. The patient's attitude was cooperative, and she quickly learned to calculate diets with such facility that she could assist the hospital dietitian in this capacity. She was discharged under good control, taking 16 units of protamine zinc insulin and 30 units of regular insulin each morning before breakfast, with diet C 225, P 80, F 75/1,895 calories, and instructed to return to the outpatient clinic for periodic recheck and guidance. Her course was uneventful through 1946-47.

An astonishing series of 35 episodes of severe coma then followed, supplemented by additional comas in other hospitals (table 1). Forty-two such episodes have actually been fully documented, and we have reason to believe there are several more not here recorded. She moved to another state in 1953

where only one episode occurred in a year and a half; then she returned to Indianapolis and was again admitted in coma in December 1954. The period 1952-53 was her championship year at Indianapolis General Hospital. It was featured by 18 admissions, an all-time record of well over one coma per month. In several instances the patient was desperately ill, having blood sugar levels ranging to 1,000 mg. per cent, extreme dehydration, and circulatory collapse. Each time recovery occurred and reasonably good control was re-established before discharge with doses of insulin ranging from 80 to 110 units daily. On each admission evidence of vascular damage and changes in ocular fundi were carefully sought but none was ever found.

Three weeks prior to the December 1954 admission to the hospital (comatose; pulse 130; blood pressure in right arm 140/80, in left arm 50/0; blood sugar 480 mg. per cent; plasma carbon dioxide 4.2 mEq./L.) careful history disclosed that she had noted the initial appearance of numbness, pallor, and coldness of the left hand and arm several months previously and had had two acute attacks. Examinations by Dr. K. R. Woolling, Department of Peripheral Vascular Diseases, established the absence of left brachial, radial, and ulnar pulsations. X ray confirmed the diagnosis of well established chronic occlusive arterial disease but did not disclose any calcification. The eye grounds remained normal. There was an obvious atrophy over the left upper extremity.

DISCUSSION

This patient is the product of a broken home. Her parents were divorced shortly before she was found to have diabetes. The mother has had eight convictions for child neglect, drunkenness, and immorality. The father was an alcoholic, her stepfather is the same: the latter has been jailed on numerous occasions for beating his wife. In 1947 the mother was sent to Woman's Prison, and it was at this time that the child began to have the tautologous comas. She was then living in a guardian's home, and this was followed by several foster homes with relatives. At any sign of friction the patient would run away and appear at the General Hospital in a few days in coma. There is evidence that the comatose state in certain instances, was deliberately induced and that it constituted a behavior pattern designed to meet an intolerable life situation. On one occasion, following a too frank discussion of this aspect of the problem by the senior author during a ward round with the resident staff, the patient left the hospital, transferred her alle-

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*Director, Medical Research Co-operation, Lilly Research Laboratories; Associate Professor of Medicine, Indiana University Medical School; Consultant, Department of Medicine, Indianapolis General Hospital.

†Resident in Medicine, Lilly Laboratories for Clinical Research, Indianapolis General Hospital.

TABLE 1
Hospital admissions

Date	Hospital	Blood sugar	Carbon dioxide mEq./L.	BUN	Blood pressure	Rabinowitch severity index	Urine acetone	Insulin first 12 hr.	Coma duration	Insulin on discharge
9/19-10/22/46	IGH	500	5	62	80/60	20	4+	500	13	51
5/26-6/8/47	IGH	435	2.7	60	120/70	11	4+	130	8½	70
8/4-9/27/47	IGH	140	22	30	120/74	<5	1+	75	3	75
10/17-10/26/47	IGH	381	6	33	118/60	12	4+	110	10	120
2/14-3/9/48	IGH	408	8.2	33	140/80	12	3+	260	11	140
6/12-6/28/48	IGH	444	13	30	130/70	7	4+	230	13	150
7/24-8/6/48	St. V.	514	—	25	120/80	>5	4+	150	—	135
9/9-9/17/48	IUMC	388	16	—	—	>7	4+	60	—	70
9/23-10/15/48	IUMC	500	20	30	—	8	4+	200	—	166
11/5-11/28/48	IUMC	520	3	—	130/70	10	4+	270	72	226
12/9-12/12/48	IUMC	400	—	—	115/90	<5	4+	60	48	226
12/23/48-1/24/49	IUMC	571	4	—	118/94	12	4+	130	24	158
2/15-3/11/49	IGH	430	4	38	126/90	12	4+	180	14	120
3/20-3/29/49	IGH	350	4	30	124/60	9	4+	260	12	130
4/4-5/17/49	IGH	512	6	34	114/78	5	4+	200	6	100
5/22-6/9/49	IGH	380	6	55	132/68	11	4+	170	17	110
6/14-6/25/49	IGH	218	4	30	130/70	13	4+	370	6	110
8/1-8/15/49	IGH	455	4	30	130/70	11	4+	300	10	160
8/18-9/9/49	IGH	500	4	—	138/80	10	4+	310	8	130
9/16-9/24/49	IGH	556	6	—	120/60	4	4+	120	7	130
10/10-10/22/49	IGH	470	4	—	118/80	5	4+	150	5	130
10/26-11/8/49	IGH	63	4	49	105/60	8	4+	80	10	115
11/14/49	IGH	37	18	—	140/70	—	neg.	0	2	—
12/6/49-1/4/50	IGH	530	4	25	134/76	12	4+	460	26	115
2/23-3/11/50	IGH	395	4	70	124/80	15	4+	340	13	140
5/20-5/27/50	IGH	525	6	—	120/70	8	4+	245	18	140
6/16-6/26/50	IGH	500	10	—	110/70	10	4+	120	11	130
7/14-7/19/50	IGH	800	12	35	130/70	5	4+	300	7	100
7/29-8/4/50	IGH	50	24	40	96/70	<5	4+	100	8	115
8/17-8/27/50	IGH	620	8	104	120/60	15	4+	80	11	115
9/22-9/29/50	IGH	50	12	29	110/90	11	4+	40	12	100
11/1-11/7/50	IGH	800	4	65	120/70	10	4+	280	11	120
11/12-11/19/50	IGH	110	11	—	100/65	5	4+	120	6	120
11/25-12/6/50	IGH	1175	5	—	100/60	14	4+	260	12	120
12/31/50-1/6/51	IGH	620	5	—	100/90	12	4+	340	16	120
2/23-3/17/51	IGH	767	4	26	135/80	9	4+	340	16	100
6/14-6/26/51	IGH	600	4	—	100/90	9	4+	160	12	120
11/29-12/21/51	IGH	740	4	21	120/70	11	4+	240	12	160
12/2-12/3/52	IGH	455	20	16	116/64	8	3+	80	8	80
2/16-2/20/53	IGH	415	8	6	115/50	9	4+	320	12	80
Out of state	B.C., Ky.	No records								
12/13-12/20/54	IGH	480	4	26	140/70	7	4+	320	8	100

giance to another institution, and there continued the repetitive course of events. Intensive psychotherapy over a period of three years was unsuccessful in effecting an adequate adjustment. An unpleasant stressful situation was met habitually by precipitation of a bout of acidosis and coma and removal from the life stress to the more sympathetic hospital environment. The patient appar-

ently committed a kind of suicide at will, with the assurance that her demise would be temporary and not permanent. Uncertainty that the usual facilities were available to bring about her restoration was followed by a period while out of the state during which only one coma occurred.

The influence of emotional states on diabetes has long

been recognized. Mirsky² presented experimental evidence that hyperglycemia can be provoked in diabetic persons during psychiatric interviews. Hinkle and associates^{3, 4} described two types of response to stress situation, one characterized by ketonemia but usually preceded by a fall in blood sugar level. If this occurs in a diabetic and is accompanied by "stress diuresis," this response may lead to ketosis. If the person is then given glucose the glucose tolerance curve is higher and more prolonged. Such a mechanism might well be the "trigger" activating the prompt development of severe diabetic comas in the present case.

The delayed development of vascular damage in spite of such dramatic persistent insults and poor control of the diabetes is also noteworthy. Approximately eight years of such tautologous behavior elapsed before positive findings occurred, and even then the eye grounds were not hemorrhagic. The long-term studies of the Joslin group, Keiding, Root, and associates^{5, 6} suggest that vascular complications are related to the degree of control of diabetes; and that under ordinary circumstances they develop in 10 to 15 years even though control may be classified as generally excellent. Our patient has provided an opportunity to observe the effect of intensive and almost constant poor control and the influence of repetitive severe comas. The fact that peripheral occlusive vascular disease has had its onset and then developed rapidly since the patient's last admission in 1953 may be of some significance.

In January 1955, after the last described admission and following a series of discussions with the patient, the junior author arranged for her employment as an assistant in the diet kitchen at Indianapolis General Hospital. Here she would undertake supervised responsibilities for the calculation of diabetic diets and could participate in teaching dietary calculations to new diabetics and those undergoing treatment in the wards of the hospital. Contact with her immediate family was discontinued, and she now lives with an aunt who has a mature understanding of the problem. The value of this psychotherapy has been reflected in the state of diabetic control, in that it is uneventfully good and has manifested only rare glycosuria over these months, with a daily dose of 60 units of insulin.

SUMMARY

A behavior syndrome termed tautologous diabetic coma and occurring in a severe juvenile diabetic person is described. It is characterized by repetitive comatose states brought about in response to disturbing and intolerable situations in the patient's life environment. Approximately fifty episodes of coma have occurred in the course

of eight years. The case is considered in relation to the effect of stressful situations on the control of diabetes and the effect of poor control on development of vascular complications. Improved social and emotional adjustment has been followed by a complete cessation of diabetic instability and comas.

SUMMARIO IN INTERLINGUA

Coma Diabetic Tautologe—Un Syndrome de Behavior: Multiple Episodios Innessari de Coma Diabetic

Es describite sub le nomine de coma diabetic tautologe un syndrome de behavior occurrente in un juvene persona con sever diabete. Le syndrome esseva caracterisate per repetitive statos comatose provocate in responsa a disturbante e intolerabile situationes in le ambiente vital del patiente. Circa 50 episodios de coma occurreva in le curso de octo annos. Nos discute iste caso in relation al effectos de situationes stressante super le domination de diabete e le effectos de inadequate domination super le disveloppamento de complicationes vascular. In le caso hic presentate, le melioration del adjustamento social e emotional resultava in le complete cessation de comas e instabilitate diabetic.

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DISCUSSIONS

H. D. BREIDAHN, M.D., (*Australia*): I should like to thank Dr. Peck for his paper and for reporting this very interesting case. I am going to make a little note about the opposite condition, because one occasionally sees diabetics with behavior problems who present hypoglycemia, self-induced. These cases have been reported—I think the first one by Dr. Rynearson—and they do present similar behavior problems. They are often very diffi-

cult to cure, and Dr. Peck's patient certainly was managed very satisfactorily. I only wish that they were all managed as well.

HENRY T. RICKETTS, M.D., (*Chicago*): We have had a patient who will run a close second to Dr. Peck's, and of a very similar nature with respect to the disturbed home background—a young girl who has been admitted 34 times in acidosis in a period of 10 years to our hospital in Chicago. Each time she would have difficulty with her family, she would refuse to eat, and then, not eating, would also omit her insulin, so that ketosis and acidosis promptly developed. As time went on, she learned that she had to take her insulin whether she ate or not, with the result that on one occasion she came into the hospital in mild acidosis, with a serum CO₂ level of 13 millimols per liter, and a blood sugar of 76 mg. per 100 cc., a perfectly normal glycemic level. This, I would suppose, is a manifestation of the capacity of emotional disturbances to produce ketosis of such a degree as to lead to frank acidosis, and probably also is an expression of the very poor glycogen reserve that this young woman had, with the result that fat metabolism was taking place at a very rapid rate. The only difference between Dr. Peck's case and ours is that we have done a better job of producing vascular lesions, perhaps, than he has. This girl now has frank micro-aneurysms in both retinae.

J.A. SMYTH, M.D., (*Northern Ireland*): In the pursuit of the biggest ever, I don't think Northern Ireland can compete with the United States of America, but we have had a patient who has been admitted 15 times in 5 years, each time in coma, and in investigating this case, it was found quite similarly that home conditions were responsible. She had no psychoneurosis as far as we could make out, and as far as our psychiatric people could advise, but she was living under wretched home conditions where life was miserable, and the bright and pleasant life in the hospital appealed to her, and when she got tired of living under these conditions, she simply stopped her insulin and was brought in in coma. There is no solution to that as yet, as far as we have been concerned, and I don't know whether we could think out something along the lines that Dr. Peck managed.

Another interesting experience occurred very early in the history of insulin, when insulin was frightfully expensive. We had a man who was admitted five times, and each time his recovery from coma cost my hospital between 400 and 500 pounds, and it presently became a problem—what were we going to do about this? Hospital finances were voluntary in those days and it was rather difficult to think of what could be done about it. How-

ever, the family practitioner solved the matter for us, because he left this man for about 36 hours in coma in his own house, and when he was brought to the hospital, we couldn't bring him out of the coma and he died.

H. WHITTAKER, M.D., (*London*): I was most interested in this report of Dr. Peck's because, of course, these are the people who, with children, come under residential care in this country, and we often get a history, even in a young child, of a series of five or seven quite severe episodes of ketosis. I am most interested in this question of emotional upset because my own feeling is that there is always something mechanically wrong, that the emotion in some way disturbs the diabetic regime.

When we have the children under residential care, emotional upset often shows itself in an attack of vomiting, and that, I think, is where the diabetes starts to go wrong. If they have a bilious attack when they are in residential care, a urine test is immediately done, and effective treatment started, and so they don't go into these severe episodes of ketosis. When they leave residential care, most of them have grown out of this behavior difficulty, but there still is a very small number—and they usually are diabetics with a rather low intelligence quotient—who will continue to give trouble in this way.

J. J. GROEN, M.D., (*Amsterdam*): We have also had several patients with diabetes in whom we have had to make a diagnosis of psychoneurosis, and very often of obvious psychopathy. Now, lately, we have come to ask ourselves how far we, as doctors, may have been responsible for the production of these abnormal mental states. At least two ways can be visualized in which these abnormal mental states can develop in people who are under medical treatment.

The first is due to the fact that many of these children when they were very young were kept in hospitals for very long periods of time. A girl we have, for instance, who has been admitted 36 times, was kept in a hospital when her diabetes was first discovered, from her third to her seventh year, because this was just about the time when insulin was brought in. She was regimented very rigidly, and she herself ascribes her present attitude toward her disease to this regimentation and imprisonment, as she calls it.

The second way in which, perhaps, we might damage the patient is through too rigid control. Sometimes they may do it themselves. By subjecting them to repeated insulin shocks, it has been demonstrated that severe, persistent hypoglycemia may lead to brain damage, and that brain damage may sometimes show itself

in psychopathic behavior disorders. Our patient went into severe hypoglycemic shocks several times, either because she didn't pay enough attention or because she was too rigidly controlled.

It has to be recognized that the real danger of damaging diabetic patients, especially young ones, can be in irregular control, both by ketosis and sometimes also, by overtreating and damaging their brain, and thereby their emotional life, via repeated severe hypoglycemia.

C. STRIKER, M.D., (*Cincinnati*): I would like to record, inasmuch as this is an experience meeting, one of our successful failures! We experienced a similar situation in an eighteen-year-old diabetic of about ten years standing, who was admitted to the hospital at least twenty times. After much psychiatric supervision and metabolic control, we admitted that we could not control the patient. We then considered the question of a prefrontal lobotomy, and it was performed. That patient now, although a transformed individual, has not been admitted to the hospital in keto-acidosis for at least two and one-half years.

H. H. BERG, M.D., (*Hamburg*): Considering the possible causes for repeated comatose diabetic states, I would ask if others have observed any influence of a seasonal factor. We have seen several cases in which coma occurred on nearly the same date, in autumn, in a sequence of some years.

DR. PECK, SR.: We have not noticed seasonal incidence in this patient of ours. In the period of 1952-53, the patient went through all seasons and had a total of eighteen comas that year.

DR. STRIKER: I apologize for rising again. I would

like to ask Dr. Peck—and all of us have had the experience—what is his explanation, if possible, of the apparent mildness of this individual after such severe trauma. One is impressed in the reversion of diabetics from their ketotic state, if the overlay of stress is removed. They almost invariably return to a relatively mild state. But here is an individual who has had many traumatic experiences and one would expect pretty severe changes in endogenous insulin supply if that is a factor. Does Dr. Peck have an explanation for the mildness after her readjustment?

DR. PECK, SR.: By mildness, you mean the insulin requirements? Well, she had been receiving between one and two hundred units for long periods of time. Since the beginning of this year, when her emotional readjustment has been so much better, she has rarely shown any glycosuria at all and has had a continuous reduction in dosage of insulin from 100 down to 60. How much farther she will go, I have no idea. I also have no good explanation of the fall that has occurred thus far. However, I do feel that the insulin requirement per se, as measured in number of units per day, does not reflect absolutely the severity, the real severity, of the diabetic in that occasionally one sees an individual who will tip over into acidosis very promptly on withdrawing a small dose of insulin, 20 or 30 units, and have ketosis within a few hours; whereas in other patients, usually the older, obese type of individual, one may see 100 units withdrawn without much particular change. I would assume that this patient, because she is young and has gone through this period of several years of traumatism, probably has a total lack of circulating insulin of her own.