SWEAT TEST FOR THE DIAGNOSIS
OF FIBROCYSTIC DISEASE OF THE PANCREAS

REPORT OF A FATALITY

BY

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(RECEIVED FOR PUBLICATION SEPTEMBER 8, 1957)

The recognition of the electrolyte disturbance in
the sweat of patients with fibrocystic disease by
Darling, diSant’ Agnese, Perera and Andersen (1953)
led Shwachman, Leubner and Catzel (1955) to devise
a simplified method of obtaining sweat for bio-
chemical assay.

In this method the patient is placed in a plastic
bag up to the neck and covered with a blanket, the
sweat being collected in a weighed square of gauze
which is covered with a piece of impermeable
sheeting and strapped to the back. The recom-
manded time is 30-90 min., depending on the rate of
sweating.

The test is now widely used. In spite of the
theoretical risks no serious ill effects from it have
been reported. We have had the unfortunate
experience of a death following its use, which we
record as a reminder of its potential danger.

Case Report

A boy, aged 13 months, was admitted to hospital on
account of failure to thrive, backwardness and diarrhoea
with bulky and offensive stools, for an unknown period.
Delivery and the neonatal period had been normal but
he sat up late, and was unable to get into a sitting position
at the time of admission. He appeared in a neglected
condition and the home conditions were bad. Apart
from his low weight, flabby musculature and slight
ichthyosis of the skin, physical examination revealed no
abnormality.

The Wassermann reaction, chest radiograph and
Mantoux test were negative, and the blood count was
normal. The stools were pale, bulky, greasy and
offensive; no pathogens were isolated. The duodenal
fluid was slightly alkaline and showed trypsin activity
to a titre of 1 : 32 (gelatin film method of Harrison,
1947). As this suggested pancreatic impairment, pan-
creatin granules were given with each meal. There was
a rapid general improvement with increase in weight
(5 lb. (2.25 g.) in four weeks) and he started to walk.

Although the child was now well, because of the
apparent pancreatic impairment a Shwachman test was
performed (after sedation with 1 ml. of paraldehyde) by
the method described above but using hot water bottles
to promote sweating. The test lasted two hours. The
result, an increase in the weight of the gauze by 0-2 g.,
with complete absence of chloride and sodium, was
equivocal, and it was decided to repeat the test a week
later. The patient was sedated with chloral hydrate
gr. 3 (0-05 g.) and, as this had no effect, paraldehyde
0·75 ml. As little sweat had been obtained in the
previous test, the period of collection was extended to
three hours under the same conditions as before.
Throughout the patient’s general condition appeared
satisfactory, though on removal from the bag he appeared
anxious and restless. This was attributed to the dis-
comforts of the test. Half an hour later he vomited
copiously. On examination then he was unconscious
and pale with a hot, moist skin, and a temperature of
104° F. (40·3° C.) in the axilla. Respirations were
irregular; pupils equal and reacting to light; deep
reflexes normal. Under treatment with tepid sponging,
subcutaneous and intravenous fluids and a mega unit
of penicillin the temperature fell to 100° F. (37·8° C.)
but the general condition deteriorated; the pupils became
temporarily unequal and deep reflexes were absent.
The pulse rate rose to 160-200 per minute. Death occurred
in coma 14 hours after conclusion of the test.

Necropsy

This was performed five and a half hours after death
and showed a well-developed boy with a somewhat rough
skin, but without external abnormalities. Throughout
the body there were scattered small and some larger
flame-shaped haemorrhages mainly involving skeletal
muscle and internal organs, especially marked on the
periarticular and left septal surfaces of the heart, the
omentum and the kidneys, and there were patches of
haemorrhage in the well-pigmented adrenals. The lungs
were uniformly and extremely congested, firm, and deep
red. The gastro-intestinal tract, liver, bile passages, pan-
creas and ducts were macroscopically normal. The spleen
was slightly enlarged (45 g.), firm, and of a deep red with sharply demarcated large Malpighian corporules. Generalized mild but not abnormal lymphoid tissue hyperplasia was noted with enlarged tonsils and adenoids and a generalized mild lymphadenopathy. The thymus was normal (24 g.).

Examination of the skull showed the meninges tense but not congested, the brain bulging with flattened convolutions and a pressure cone of the cerebellum. A small grey nodule, 3-4 mm. in diameter, was noted projecting from the tuber cinereum close to the infundibulum to the left of the midline. Sectioning of the brain (1,200 g.) showed dry, pale, bulging surfaces suggesting interstitial oedema. A small petechial haemorrhage was seen in the floor of the fourth ventricle. No further nodules were found.

Histology

Sections of the pancreas revealed no evidence of fibrocystic disease, and serial blocks of the terminal centimetre of the main duct showed no obstruction. The acinar cells were filled with secreting granules. The lungs showed extreme congestion only, with no abnormality of structure. The liver was normal apart from very scant round-cell infiltration of the portal tracts. Tonsils, cervical and abdominal lymph nodes, spleen and intestine confirmed slight generalized non-specific hyperplasia of the lymphoid tissue. No sign of an inflammatory reaction was seen in brain and meninges. Sections of cerebral cortex, corpus striatum and hypothalamus confirmed the oedema and showed a few small scattered capillary haemorrhages. The nodule on the tuber cinereum was a small lipoma of the pia mater. The abdominal skin showed hyperkeratosis, deficiency of the stratum granulosum and normal but inactive sweat glands (lumina closed).

Bacteriology

Films and cultures of the cerebrospinal fluid and spleen revealed no micro-organisms.

Pathological Summary


Comment

The necropsy findings, cerebral oedema and scattered body haemorrhages, taken in conjunction with the sequence of events, bears out the diagnosis of heat stroke, although the highest body temperature recorded, the first to be taken, and when the collapse had already occurred, was only 104° F. (40-3°C.) in the groin.

The method of obtaining sweat as devised by Shwachman is a simple and most useful diagnostic procedure but the potential danger of interference with the normal heat regulation of the body ought to be clearly recognized. At an external temperature of over 90°F. (32-2°C.) evaporation of sweat is almost the sole means of maintaining the normal body temperature (Best and Taylor, 1955). In the original experiments by Darling et al. (1953) a room heated to 90°F. was used to promote sweating, but in the Shwachman test the enclosure of the body by a plastic bag prevents any evaporation and permits a dangerous hyperpyrexia to occur. Patients with fibrocystic disease are known to sweat excessively and the danger of heat exhaustion due to excessive loss of electrolytes has been recorded by Kessler and Andersen (1951). This, however, need not be considered here as it bears no relation to hyperthermia. It is of interest though that the only fatality in their series occurred in an infant which was brought to the hospital during a heat wave wrapped up in a thick woollen blanket, thus preventing heat loss, and having a temperature of 106°F. (41°C.).

The simple screening test of Shwachman and Gahn (1956) which merely requires the placing of a hand on an agar plate incorporating silver chloride and an indicator, might be used more widely to demonstrate the increase of chloride in sweat of patients with fibrocystic disease. In its interpretation the possibility of a doubtful result ought to be borne in mind. We have found one false negative in a severely ill child in which fibrocystic disease of the pancreas was confirmed at necropsy. The polythene bag test should be limited to doubtful cases and when performed due regard must be given to the possibility of hyperthermia, and frequent recordings must be made of the body temperature. The duration of the test should be limited to the time specified by Shwachman and additional heat by hot water bottles or electric blankets should be avoided.

Summary

A fatality due to heat stroke following Shwachman's test involving the collection of sweat for the diagnosis of fibrocystic disease is reported. The potential danger of the test is emphasized.

We wish to thank Dr. C. G. Fagg at whose suggestion this case is being reported, Dr. H. Haber for his advice on the skin sections, and H.M. Coroner, Mr. J. H. Hoare, for permission to publish.

REFERENCES