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AN UNUSUAL CASE OF CYANOSIS.

In January, 1902, I was called hurriedly to see an unmarried woman who was very ill. I found her in bed, a well-nourished woman of 39, extremely cyanosed, almost hideous in colour, with black lips, leaden-coloured skin and blue-black nails. She was not, however, so distressed as her colour promised. There was no orthopnoea. She was lying slightly propped up in bed, breathing quietly and shallowly, with long pauses between the respirations. She was mentally quite alert and could carry on conversation in short sentences.

The pulse was of fair volume, regular, soft, and compressible, beating 100 per minute. The vessel wall was normal to touch. There was not the slightest oedema of feet or legs.

The cardiac impulse was feeble and diffuse, reaching to 1" beyond the nipple in the 4th interspace. There was epigastric pulsation. The cardiac-dulness extended to the right border of the sternium the 3rd rib, and for 1" beyond the nipple. There was a marked V.S. murmur audible over the whole cardiac area and indeed the whole chest, but more marked over the apex and towards the axilla. An occasional moist rale behind was the only abnormality in the lungs. Percussion was resonant. R.M. V.F. and V.R. all were normal.

Liver dulness was normal.

In the abdomen she complained of great tenderness over the course of the colon but there was nothing else, no distension, no unusual percussion, dulness /

dulness, no palpability of the kidneys, no evidence of enlargement of the spleen. There were no urinary symptoms, and the urine, which was examined later, was quite normal, of a lager beer colour, acid, free from albumen, sugar, and casts. There were no abnormal pelvic symptoms.

I was told that the attack had begun about an hour before and would pass off in another hour or two, leaving her quite well, and that she had been subject to such seizures for over a year. Her people were quite out of sympathy with her. She had been taking hysterical attacks for 15 years, and they thought these another form of the same. Her mother accused her of blackening her face with soot and altogether they contradicted each other so freely and so bitterly that I was compelled to believe only what I could see.

In a few hours the cyanosis was less and next day it had quite disappeared, leaving her with a sallow and unhealthy complexion which I found to be usual to her. She had coughed up a little blood-stained mucus after the attack passed, and there was still an occasional rale in the chest. The pulse was normal in rate and character. The cardiac dilatation and the murmur less distinct, and by another day the heart to the most careful examination seemed absolutely normal in rate, impulse, size, and sounds, while the lungs were perfectly clear.

A few days later there was another attack four

or five hours in length, precisely like the first, and after that for two years a long series of attacks differing in severity and at intervals varying, without apparent reason, from a day or two to some weeks.

Meanwhile, I was able to piece together her history to my own satisfaction. It was necessary to do so very securely because she was the cause of much family ill-feeling. Her people were so prejudiced against her that their statements, though made in perfect good faith, had to be considered very sceptically, and the patient's manner was so obnoxious that it was difficult not to be impatient of her. But by collating her own story, my own observations and those of her former medical attendant, with the accounts of her relatives and advising neighbours, it was not difficult to form a reliable picture.

She had been an exceedingly healthy baby and child, with no suspicion of cardiac abnormality. The only suggestion of circulatory disturbance was the fact that she had been advised to give up sea-bathing in girlhood on account of its causing a condition of 'white fingers' from 'failure of the circulation through them.' But for 15 years, from an attack of 'gastric fever' at the age of 24, she had been a family plague, taking no share of the burden of the house, considering herself an invalid and giving way to repeated 'hysterical' attacks. The 'hysterical' condition had never gone to greater lengths than these attacks, however. She had had no complaint of anaesthesia or contractures or paralysis of any kind. While I knew her she occupied /

occupied a bedroom to the front of the house where she lay in state, with a sharp eye for any visitor who might enter at the front gate. Here she received her guests and entertained them with her tale of woe, and to this room she compelled her aged and rheumatic mother to carry her meals.

Through the open window she sent messages to the village by means of grocers' or butchers' boys, and by them she received whatever she cared to order for herself and this included a considerable quant--ity of various patent and other medicines. Through the day she lay in bed and demanded attention. If she rose she did not dress completely, but preferred to appear as the invalid in dressing gown. At night she built the fire higher, ran about the house, cooked or washed as she chose, and wrote her letters or the sentimental doggerel she was addicted to. Once every month or two she rose fairly early in the morning and went off on business to Glasgow where she tripped about for many hours, returning in the afternoon to take again to bed and complain of ex-haustion. She refused to go out or take exercise or to be controlled in any way unless through fear of being left.

Apart from the attacks of cyanosis she seemed healthy in every way. She complained constantly of alternating diarrhoea and constipation, but for this there was cause enough, for she would not use laxative medicine, and would eat only hot and spicy foods, and even these were gulped down at a great rate. She was never without a pain over the stomach or the head, the heart, the manubrium, the lungs /

lungs, the back, and the pain was always agonising, whether it was referred to an acne-pustule of the face, which she would call lupus, or to some undiscovered cancer of the stomach, which she was certain was hurrying her to her grave.

But apart from the attacks nothing more was to be found. She was well-nourished and fairly well-coloured, and though sallow without the slightest lividity. There was no clubbing of the finger tips or curving of the nails, and no oedema of feet or ankles. She was never feverish.
There was no cough, no breathlessness, never even mild bronchitis, only the few moist rales after an attack, and no other abnormality in the lungs.
The heart between the attacks seemed normal in every way. By and bye when I could get her to walk half a mile to see me she seemed only the better for it and no heart murmur or dilatation appeared even after the exhaustion of a day in town.
The blood vessels were soft and healthy even to the retina.
There were no urinary symptoms, no increase in the daily quanitity, no albumen, or sugar or casts.
There was no enlargement of spleen.
Repeated careful examinations revealed no abnormality in any part. The blood was not examined.

The attacks varied in duration and severity, and in number without apparent reason. They seemed rather less numerous before her visits to Glasgow than after, and especially so if the visits were postponed for a month. They were certainly not brought on

by /

by exertion or by anger. They lasted from half an hour up to 4 or 5 hours when very severe. The cyanosis was always by far the most prominent symptom and out of all proportion to the other symptoms. Her lips were not blue but black, and the skin generally of a slate grey or leaden tint. But of other distress there was little. She had none of the deep painful breathing we associate with cyanosis in ordinary heart cases of sudden heart failure. She could lie down and talk with comparative ease. The respirations were indeed remarkably quiet and shallow and the pause much prolonged. She did not give the impression of holding the breath. There was not the swollen congested face of asphyxia. Deep voluntary respirations did not lessen the cyanosis in the least, but a period of still shallower breathing followed like the healthy "apnoea" that normally follows the same cause.

The cardiac condition in the attacks was apparently that of dilatation of both sides of the heart. The diffuse pulsation extending for at least 1" beyond the nipple (the breast was small) and the V.S. murmur where both very distinct and both disappeared within two days of an attack.

Treatment was useless. All the usual cardiac and other tonics were tried without apparent influence. She had an infinite capacity for taking medicine and no want of faith in it, but objective improvement could not be obtained.
She could only be managed by her fear of being abandoned, and by means of this she was got out of bed and out of doors. The attacks, though irregular, came /

came less often. Gradually as my visits became less frequent but regular, her mother began to assert that the attacks always came on when the doctor was expected. As she had at first accused her daughter of blackening her face with soot or dye of some sort this assertion was not received without scepticism. By means of unexpected visits and with the help of the district nurse, who was allowed to call, to massage the patient, it was slowly but completely corroborated, and then the cure was rapid. It was explained to the patient that while in the attacks her life was in danger, that if she stopped the use of drugs she would be free from attacks and that she would receive no attention during future attacks unless at her mother's request. The attacks ceased, almost immediately and she has remained well since, except for one attack some months later, which occurred while I was on holiday but which did not recur when she found that the locum tenens had been forewarned.

The first question that must be considered is this case, is the nature and cause of the attacks.

About the cyanosis there can be no doubt. Severe cyanosis is a condition than cannot be mistaken.

That the attacks were caused by no abnormality of circulatory or respiratory systems, there seemed to be no doubt either. Repeated careful examinations between the attacks, both during rest and after rather severe exertion, revealed no lesion in either side of the heart or in bloodvessels, kidneys, or lungs, and what is perhaps equally convincing, the attacks were /

were never caused by even prolonged exertion, for, on the contrary, that only brought a healthy red glow to her cheeks, hiding the somewhat sallow colour that was more usual to her. No doubt a patent Foramen Ovale, or even a more serious congenital abnormality may be present in a heart that to ordinary examination seems quite normal, but even if present, such a condition would not explain the independence of the cyanosis of cardiac strain. The healthy state of her early years gives no support to such a suggestion.

The attacks again seemed equally certainly to be under control. It was necessary for many reasons to make sure of this. The domestic atmosphere was one of discord. The patient had 'cried wolf' so often that when I saw her her relatives at their kindest simply ignored her and resented my presence. But even a hysterical patient must die once, and her death during an attack would no doubt have completely reversed their attitude. She could not afford to go to a Nursing Home or even to a consultant, and it was impossible to watch her. The conclusion that she brought on the attacks was only reached after every fallacy served to be excluded. That it was correct was almost proved by the immediate cessation of the attacks and still more by the solitary attack that occurred some months later when a stranger was certain to attend her.

Cyanosis that is under control may have several possible explanations but only one that is probable. The heart is not under voluntary control. A few doubtful cases, such as that of Colonel Townsend, mentioned in Taylor's "Forensic Medicine" and some

cases /

cases mentioned in Ogston's "Medical Jurisprudence" only emphasize the fact. These occurred in subjects of serious disease and partook of the nature of syncopal attacks.

Hysteria is a disease of protean vasomoter symptoms. These, if we cast the net widely enough, are capable of producing a local cyanosis, the local disturbance not appreciably affecting the general circulation. It is difficult, however, to imagine the whole body in a state bordering on inflammation without very serious results. It is still more difficult to imagine this being produced voluntarily. A similar disturbance of the nervous control of the heart rather than the vessels might cause such cyanosis. But again, this is contrary to all experience In the presence of a simple explanation it is almost useless to discuss it. The hysteria of the present case too had been merely a psychical one, without any of these more serious vasomotor changes. The ' 'dead fingers' from which she suffered after sea-bathing, prove nothing.

The question of whether cyanosis can be produced voluntarily by holding the breath is more difficult to decide. In the present case it was considered frequently during the attacks. Her comparatively comfortable appearance, and above all, the failure of deep voluntary respirations to lessen the cyanosis seemed to negative this explanation.

The explanation which seemed the only reasonable one and seemed further to be justified by results was that the cyanosis was produced by the use of drugs. True, the patient would not admit it when asked point-blank /

blank about it months later when she was well, but then she was always untruthful when it suited her. Probably such a confession was too much to expect of human nature, especially in a hysterical form. Certainly she was never happy unless when swallowing medicine of some kind, and if none were prescribed she supplied herself with a constant stream of headache powders, patent medicines, pills, tabloids, and stuff of all kinds. She was willing to admit the use of Chlorodyne in small doses and of some homeopathic pellets of Digitalis and Cactus, so that probably these were not the cause of the cyanosis. Certainly she was never sleepy or even heavy during the attacks, but on the contrary quite alert.

Many drugs cause cyanosis among the later symptoms of serious poisoning, but few are credited with the power of producing it as the only symptom of any consequence. It was considered that the cyanosis was due most probably to one of the modern coal-tar analgesics, both because these are powerful cardiac depressants and with little obvious influence elsewhere, and because they are widely sold to the general public and most likely to be used freely by a woman always complaining of neuralgia and headache. The rapid cure following a pointed explanation almost clinched the diagnosis, and my confidence in it was not diminished by finding later that Osler describes a similar case.

Reported cases of cyanosis independent of disease of heart or lungs are not numerous.

In the Lancet of 1886 Rayner described a number /

number of cases of deep cyanosis in children a few days or weeks old, from the absorption of Aniline from a marking ink used for their napkins. The Aniline was absorbed in 24 hours or excreted in 5 days. The depth of the cyanosis and the slightness of the other symptoms were the most marked clinical features.

In the Lancet *Feb 22 - 1902* Saundby & Russell described three cases of cyanosis with enlarged spleen, and these were incorporated with three cases of his own and some others in a paper in Brit. Med. Journal, Jan. 16, 1904, by Osler on Chronic Cyanotic Polycythaemia with enlarged spleen. In these cases all were cyanosed without respiratory distress and without obvious cause. Advanced emphysema and chronic heart disease could be excluded. Some of the patients enjoyed good health in spite of extreme cyanosis, one woman being known as a curiosity in the countryside, while carrying on her daily work. In other cases prostration was great and death ensued. The blood was dark and very viscid and in 8 cases contained above 9,000,000 Red Corpuscles per cub. Mm. with 120-150% of haemoglobin, while the leucocytes were uncharged. In 7 out of 9 cases the spleen was much enlarged, reaching the navel in 4. There was pigmentation of the skin in 5 cases.

Osler mentioned two cases of tubercle of the spleen with similar symptoms.

He mentioned also the possibility of chronic poisoning with coal-tar products and in particular a case of chronic cyanosis of two years' duration, without splenic enlargement or hyperglobulisin, where the taking of Acetanilide was finally discovered though /

though till then denied, and also another reported case of acute Acetanilide poisoning with extreme cyanosis and a spleen palpable for 2" below the ribs.

Whitla mentions that many cases have been reported of poisoning from even 5 qrs. of Antifebrine with cyanosis and extreme collapse.

Reviewing this case two points seem noteworthy. The first is the cause of the cyanosis. The shallow character of the respirations, contrasting so vividly with the orthopnoea of 'cardiac cases' points probably to some depression of the respiratory centre which falls into line with the cerebral depressant action which makes Analgesics of Antipyrin and its class. How far the circulatory depression was due to a similar action on the cardiac ganglia and the Pneumo-gastric centre, and how far to a direct poisoning of the cardiac muscle is a question which probably cannot be answered.

But cardiac and respiratory depression alone scarcely explain the depth of the cyanosis. A soft regular pulse of 100 not feeble enough to cause immediate anxiety, even in the presence of distinct signs of cardiac dilatation does not accord with a cyanosis which renders the lips and tongue black. Antifebrine is large doses breaks up the red blood corpuscles in numbers forming Methaemoglobin. In the present case the attacks were not followed by any serious anaemia. Short of this it possibly forms with Haemoglobin a temporary compound analogous to Carbon-Monoxide-Haemoglobin but darker than Oxyhaemoglobin and so capable of giving a darker colour /

colour to the blood. The possibility of cyanosis from the formation of dark Haemoglobin compounds should be remembered in considering the association of cyanosis with enlargement of the spleen.

The second point is the contrast between the depth of the cyanosis and the comparative slightness of other distress. This contrast has been noted in many conditions. It was present in the cases of Aniline poisoning and in some of Osler's cases and it has been the cause of much discussion in connection with congenital heart disease.

Much of the difficulty must arise from the fact that our standard for degree and distress in cyanosis is set by "ordinary cardiac cases" where cyanosis is one of the terminal phenomena, the patient before cyanosis develops, being generally at the point of death, and indeed in many cases dying solely from the earlier consequences of circulatory failure.

Even in cases of congenital malformation of the heart this consideration has some bearing.

In spite of the variety of the pathological conditions that is found in such cases, the effect on the circulation viewed broadly is similar in almost every case, in one respect, that the aeration of the blood is carried on by the passage of a portion only of the general blood stream through the lungs, and so as the circulation in such cases gradually fails the symptoms due to defective aeration must take an earlier and more prominent place in the succession of consequences.

In the present case there was no reason to think that there was any congenital abnormality to assist in the production of cyanosis. Even a hypothetic--ally patent foramen-ovale could not help. There have//

have been cases where a patent Foramen-Ovale found post-mortem was the only explanation for frequent or constant cyanosis during life, and others where the same condition had been preceded by no symptoms of any kind. To explain the contrast we must suppose in the first case some obstruction to the pulmonary circulation, which by raising the blood pressure in the right auricle led to a short circuiting of part of the blood stream through the Foramen-Ovale, but in the present case there was no Bronchitis and no other cause of obstruction of the pulmonic circulation.

Summarising we might describe the case as one of intermittent and severe Cyanosis, induced almost certainly by drugs, in a healthy but hysterical woman, with special interest from its bearing on the physical causes of Cyanosis.

John Crow

Largs