A Case of Megalosigmoid with Incontinentia Alvi; and the Nuclear Picture.*

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By

Hosaku Namekawa.

(滑川方策)

(From the Department of Pediatrics, Faculty of Medicine,
the Tohoku Imperial University, Sendai.

Director: Prof. A. Sato.)

Introduction.

In 1886 Hirschsprung¹⁾ first described megacolon under the title of "Stuhlträgheit Neugeborener in Folge von Dilatation und Hypertrophie des Kolon". Since then numerous cases of idiopathic dilatation of the colon have been reported as "Hirschsprung's disease".

This remarkable condition is characterised by profound constipation and enormous distension of the abdomen, due to gross dilatation and hypertrophy of the colon. But cases accompanied with incontinentia alvi are relatively scarce: within my knowledge 1 case was reported by Hiro²⁾, 3 cases by Chaudhuri³⁾, 4 cases by Hallé⁴⁾, 3 cases by Hug⁴⁾, 1 case by Terrien⁴⁾ and 1 case by Shirono⁵⁾. Takats and Biggs⁶⁾ reported the following one case. In a patient an imperforate anus was operated after birth and he developed fair control of his bowels until November in 4 years, when he had no bowel movements for ten days. After several cathartics were given, he developed a fecal incontinence. In this case megacolon was, Takats and Biggs stated, detected by X-ray examination.

My own patient was admitted to our Clinic with the complaint of incontinentia alvi, and the diagnosis of Hirschsprung's disease was made only by X-ray examination.

Report of Case.

S.F., male aged 7 years and 8 months, was admitted in Jan. 29, 1939. Family and past history were of no significance.

History of the present disease :—In early childhood he never excreted stool involuntarily. In summer of the age of 4 years he suffered from dyspepsia and recovered after 2 weeks. Since then he had had incontinentia alvi, but no enuresis nocturna. The stool was, as it was stated, a dark brown clot and was found whenever napkins were changed. And the amount of stool was, as mother stated, always small, even if an enema or a laxative was used.

Examination on admission :—His constitution was rather thin and delicate. Intelligence was developed normally. The throat was not injected. The lymph nodes of cervical and submaxillary region were not enlarged. Dermographism was negative. Heart and lungs were normal. No pathologic finding in urin.

The abdomen was not so distended and dilatation of epigastric veins and intestinal peristalsis were not found. The abdominal reflex was rather weak. On the left hypogastric region, an intestinal tumor like mass was palpable which was slightly painful, removable with finger pressure. This region was tympanitic, somewhat dull. Surroundings of anus were stained, with faeces. On rectal examination, anal sphincter was normal and no stricture was found. The stool was corporeal and like the excret of a rabbit.

Fig. Megalosigmoid.

From X-ray examination the diagnosis of Hirschsprung's disease was confirmed (Cf. X-ray photo and Fig.). This case was not such a one as ll make the diagnosis probable on the very first inspection. A barium enema

showed the remarkable dilatation and the elongation of sigmoid colon and rectum. And when a barium meal was given perorally, it became clear that the stomach and the small intestines were normal, while there was a retention of barium in the large intestines, especially in the under part of them.

The course of disease :—During admission he had rarely slight pain and distension of abdomen.

Nakajima⁷⁾ reported that in his case of incontinentia alvi he used yakriton in an amount of 0.5 c.c. (probably 1/2 R.A.U.) every day for 5 days and that he was able to cure the case completely, but in my present case yakriton was not effective, although this was injected in an amount of 1 R.A.U. every day for longer than a month.

TABLE
Blood Picture of Megalosigmoid.

Day of examination			29. I	18. II	7. III
Red cell count per cmm. (million)			510	500	483
Hemoglobin (Sahli's value)			85	85	80
White cell count per cmm.			10,650	7,000	8,300
Varieties of leucocytes	Basophiles	Absolute number (%)	30 (0.3)	60 (0.8)	80 (1.0)
	Eosinophiles	"	230 (2.2)	180 (2.5)	190 (2.3)
	Neutrophiles	"	4,050 (38.0)	3,260 (46.5)	4,020 (48.4)
	Monocytes	"	800 (7.5)	390 (5.7)	440 (5.3)
	Lymphocytes	"	5,540 (52.0)	3,110 (44.5)	3,570 (43.0)
Arneth's nuclear shift	I	M	0	0	0
		W	0	0	0
		T	3.8	6.0	6.5
	II		34.2	30.0	45.5
	III		41.0	52.0	42.5
	IV		21.0	10.3	5.0
Nuclear picture of neutrophiles	V		0	1.7	0.5
	Semicolon-typed		0.5	0.7	1.0
	Comma-typed		5	7	4
	C-typed		31	22	30
Sedimentation of red cells (Yoshida's micro method)	O-typed		64	71	66
	15'		0.5	0.5	1.0
	30'		1.0	1.5	2.0
	45'		2.0	2.5	4.0
	60'		2.5	3.5	5.0

In spite of our advice for an operation, his family did not permit it and he was discharged from our Hospital in almost the same condition as at the admission.

Blood Examination.

Blood was examined three times (19.I, 18.II, 7.III, 1939) using the Tohoku-Pediatric Method⁸⁾, a modification of Sato and Sekiya's copper peroxidase reaction⁹⁾.

In the 1st examination leucocytosis resulting from lymphocytosis and monocytosis was seen, but generally speaking, in all the blood pictures and also in hemosedimentation, there was no pathologic findings. My interest was, however, the nuclear picture.

In 1933, Hayes, Spézin and Oransky¹⁰⁾ reported that the topical diagnosis of a disease could be made from the study of nuclear forms of neutrophile leucocytes of blood films. Suzuki¹¹⁾ of our Department gave the names: O-typed, C-typed, Comma-typed and Semicolon typed cells. If their opinion is expressed in Suzuki's terms, O-typed cells will be dominant in affections of the undermost intestinal part below flexura sigmoidea, C-typed cells in affections of colon descendens, transversum or ascendens and of appendix, the Comma-typed in affections of small intestines, omentum, bile passages, subphrenic space and pleura and Semicolon typed cells in pulmonary affections. It will not be difficult to presume the predominance of O-typed neutrophiles in my own case. And the actual result was as had been expected (Cf. Table of Blood Picture).

But this coincidence needs an explanation. In the opinion of Hayes, Spézin and Oransky, O-typed cells will become predominant in a case of obstipation or diarrhoea. My own case, being a case of megacolon, may be a case of obstipation in a sense. The predominance of O-typed cells due to the obstipation cannot be wholly excluded.

Summary.

Shirono⁵⁾ reported one case of Hirschsprung's disease in cases of encopresis, and he stated that it was a question whether incontinentia alvi was a symptom of Hirschsprung's disease or Hirschsprung's disease was accompanied with encopresis. My own case of Hirschsprung's disease was accompanied with encopresis too. My impression is: American cases are not accompanied by encopresis (Cf. almost all of the 9 cases of Takats and Biggs⁶⁾), while in French cases the con-comitance is not rare (Cf. Hallé, Hug and Terrien⁴⁾).

Concerning blood picture, Hiro¹²⁾ stated that there was no remarkable difference from normal blood picture, but that the nuclei of neutrophiles shifted more or less to the left. In my own case blood examinations were performed three times, but showed normal blood pictures, except for a leucocytosis on the first examination and for the nuclear picture above mentioned.

According to Hayes, Spézin and Oransky, Suzuki and Shindo, Kokubo and Kimura¹³⁾ the appearance of the cells with O-typed nucleus in a dominant number showed the affection of the intestinal canal below flexura sigmoidea. In my own case it was very remarkable, for the cells with O-typed nucleus appeared in the highest percentage among the staff cells.

Conclusion.

I reported a case of Hirschsprung's disease with the symptom of incontinentia alvi with blood pictures. In nuclear picture cells with O-typed nucleus were dominant among the staff neutrophiles.

REFERENCES.

* Read at the 26th Meeting of the Sendai Branch of the Japanese Pediatric Society, held on September 30, 1939.

- 1) H. Hirschsprung, Jahrb. f. Kinderhk., 1888, 27, 1.
 - 2) Y. Hiro, Jika Zassi (Acta Pediatr. Jap.), 1927, No. 321, 226.
 - 3) C. Chaudhuri, Arch. f. Kinderheilk, 1932, 97, 29.
 - 4) Discussion of M. Heuyer and Mme Roudinesco, Bull. de la soc. de Pediatr. de Paris, 1937, No. 10, 704.
 - 5) F. Shirono, Jika Shinryo, 1939, 5, 644.
 - 6) G. de Takats and A.D. Biggs, J. Pediatr., 1938, 13, 819.
 - 7) K. Nakajima, Chiryo-Yakuho, 1939, No. 443, 11.
 - 8) A. Sato, T. Suzuki and Ry. Shibata, Tohoku J. Exp. Med., 1934, 24, 195.
 - 9) A. Sato and S. Sekiya, Tohoku J. Exp. Med., 1926, 7, 111.
 - 10) I. A. Hayes, A. Spézin and P. Oransky, Arch. f. kl. Chir., 1933, 175, 300.
 - 11) T. Suzuki, Tohoku J. Exp. Med., 1936, 29, 108.
 - 12) M. Shindo, Y. Kokubo and S. Kimura, Tohoku J. Exp. Med., 1938, 33, 304.
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