A CASE OF HUGE PENILE AND SCROTAL ELEPHANTIASIS

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Abstract: From 1955 through 1970 the transition of filarial symptomatic patients in a town of Bonotsu, located on the southernmost part of Satsuma peninsular, Kagoshima prefecture, had been investigated. The investigations showed that number of hydrocele and chyluria patients had a tendency of decrease, though new such cases occurred at times. New cases of elephantiasis were not found as far as our investigations were concerned and the number of the disease reduced by half in ten years. A case of huge elephantiasis of the scrotum and the penis we saw lately is described. The patient is a male, 44 years old, who lives in Tokunoshima, Oshima-gun, Kagoshima prefecture. The first filarial fever attack was noticed when he was 14 years old and since then the attack would usually occur after labor once or twice a month. The patient's penis had started to increase in size gradually when he was 17 years old and at 34 it grew as low as his knee joints. Afterwards the patient's penis has grown as long as his legs without being accompanied by episodes of fever attacks or chyluria. The patient's waist is 66.3 cm (circumference) and the largest part of the scrotum 76.8 cm (circumference), which shows the scrotum is larger than the patient's waist by more than 10.0 cm. The length of the penis is 58.0 cm; the smallest part, coronary sulcus, 43.5 cm. A circumference of the glans penis is 50.5 cm. Total weight of the penis and the scrotum amounts to about 18.5 kg, which forms one-third the body weight, in contrast with the patient's height (142.0 cm) and body weight (52.6 kg). We report scrotal elephantiasis which is accompanied by penile elephantiasis, that is thought to be very rare.

INTRODUCTION

Filariasis presents such variegated symptoms as filarial fever attack, lymphadenovary, chyluria, hydrocele and elephantiasis, etc..

Among those abnormal symptoms elephantiasis has drawn attention of the public from old times due to its unusual shape. In Japan The Kumagaishi — a historical book written about Kuma (Hitoyoshi, Kumamoto prefecture) district — describes hydrocele that was seen as far back as the days of the 24th, Haruhiro (1512-1555), of the Sagara family and a famous painter, Hokusai Katsushika (1760-1849) painted an ukiyoe — a picture of everyday life in old Japan — of scrotal elephantiasis, which facts may indicate that in Japan filariasis has been prevalent from old times.

Typical elephantiasis cases, however, have rarely been seen recently. Table I

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shows results of follow-up studies on filarial symptomatic patients made by the authors for 16 years (1954–1970) at Kiyohara which has about 1,000 population, Bonotsu-cho located on the southernmost part of Satsuma peninsular, Kagoshima prefecture. Bancroftian microfilaria positive rate among inhabitants of Kiyohara was 8.6% in 1962.

There had been eleven elephantiasis patients in 1954 and thereafter until 1965 four of them died (cause unknown) and one moved out, which made six patients were still living on investigation in 1970. Any newly occurred elephantiasis case was not found during 1954–1970. In 1954 there had been twenty-seven hydrocele patients and thereafter nine of them died and one moved out till 1965 — decrease of ten hydrocele patients —, though three new cases occurred during the period. Later in 1970 another new case of hydrocele was found.

Fourteen chyluria patients had been confirmed by the authors in 1954 and on the occasion of investigation made in 1965 it was found that three of them died, one moved out and seven new cases occurred. In addition, two new cases of chyluria were also found in 1970.

As shown above, new chyluria cases have been found even recently, though, on the contrary, elephantiasis and hydrocele patients have become relatively rare in occurrence these days.

A case of huge penile and scrotal elephantiasis we have found lately is presented in this paper.

CASE

- 1) The patient (K.H.): 44 years old, male.
- 2) Present Address: Tokunoshima (Toku island), Ohshima-gun, Kagoshima prefecture.
- 3) Chief Complaints: Scrotal and penile enlargement, swelling of left lower extremity.
- 4) Family History: Non-contributory.

Past History: The patient has been of inborn mental deficiency to the extent that his mental power could not be expressed by the I.Q. (intelligent quotient), which forced him to give up school after first grader. A diagnosis of hypertension was made on the patient about two months ago.

Present Illness: He had lived in a hut in mountaneous terrain until thirteen years old and had never used a mosquito net notwithstanding lots of mosquitoes in and around the area. After thirteen year old he has lived in a sparsely-populated village in the level land. A filarial fever attack (high fever, ague and shiver) started to develop once or twice a month when he was fourteen years old, which continued until thirty. The fever attack usually occurred after labor such as mowing. His penis had begun to increase in size gradually at the age of seventeen and until thirty-four it grew as low as his knee joint. At the age of thirty-four microfilariae were detected in the peripheral blood by mass blood survey performed by the health center. Diethylcarbamazine administration to him was instituted, which was suspended halfway later due to an unknown cause. Blood examination for microfilaria detection

made on him at the age of forty showed negative. An episode of chyluria has never been seen with him so far. No any episode of a fever attack has been noted recently, either. His penis at present has grown as long as his lower extremity. The patient has never complained of dysuria except 'oozing out of urine from the tip of the penis' when he feels urination terminates.

System Review: Physique moderately developed. Nourishment fair. Countenance is idiotic with a relatively smaller head in size. Puls rate is 74 per minute with regular rythm and good tension. Blood pressure 152/70 mmHg. Neither anaemia nor icterus are noted. Physical findings of the lungs, the cardiac and the abdomen are not remarkable. There is no significant lymphadenopathy in the cervical and the axillary regions. A soybean-sized lymph node is palpable in the both inguinal regions. There is overall enlargement of the left lower extremity but neither redness nor keratosis of the skin are noted. The huge penis and scrotum are seen.

Findings of the scrotum and the penis (Fig. 1 and photographs):

Measurements are as follows: Total weight of both the penis and the scrotum

Fig. 1 Measurements (cm).

Body length 142.0 cm; Body weight 52.6 kg; Penis+Scrotum 18.5 kg

TABLE 1 Transition of symptomatic patients at Kiyohara

Diseases	Transition of No. of symptomatic patients in 11 years					as of 1965	newly-occurred cases in 1970
	1954	died patients	moved out patients	total	newly-occurred patients		
Elephantiasis	11	4	1	5	0	6	0
Hydrocele	27	9	1	10	3	20	1
Chyluria	14	3	1	4	7	17	2
Total	52	16	3	19	10	43	3

amounts to about 18.5 kg, which forms one-third the body weight, in contrast with his height (142.0 cm) and body weight (52.6 kg). The length of the penis is 58.0 cm. The largest part of the scrotum measures 76.8 cm in circumference which is larger than the waist (66.3 cm) by more than 10.0 cm. The smallest part of the penis, coronary sulcus, is 43.5 cm and glans penis 50.5 cm. A 'cleft' is visualized on the tip of the penis, out of which urine is seen oozed, though an external meatus urinarius cannot be confirmed. The surface of the penis is rigid and uneven with abundant warty and nodular protuberances and furrows on it. Scattered brown or black pigmentary deposits are seen as well. Elephantiasis of the left lower extremity is present, but keratosis of its skin is not so remarkable.

Laboratory Data (Table 2): The main laboratory data are as follows:

TABLE 2 Results of laboratory examination

1. Microfilaria negative		Albumin	48
2. FPT Intradermal reaction positive		BUN	20 mg/dl
3. VBS Intradermal reaction negative		Uric acid	7.0 mg/dl
4. Blood examination		Creatinine	0.7 mg/dl
Hb	13.4 g/dl	Na	132.7 mEq/l
RBC	389×10^4	K	4.7 mEq/l
WBC	8,250	Cl	105.0 mEq/l
5. Blood picture		Ca	4.15 mEq/l
Stab	3%	P	4.4 mg/dl
Seg I	6	Fe	120.0 mcg/dl
Seg II	18	Icterus index	4
Seg III	20	SGOT	18
Eosino	5	SGPT	9
Baso	1	TTT	2.9
Lymph	44	ZTT	11.0
Mono	3	Cholesterol	170 mg/dl
6. BSR		Ester ratio	78.7%
30 min. 10 mm		Lipoprotein	388 mg/dl
60 min. 27 mm		Neutral fat	125 mg/dl
120 min. 58 mm		Phospholipid	140 mg/dl
7. Urinalysis		NEFA	0.41 mg/dl
pH	6.0	Alkaline Phosphate	6.0 u
Protein	(+)	Choline esterase	0.68
Sugar	(-)	Amylase	170 u
Urobilinogen	(±)	LAP	85 u
Keton body	(-)	Au Antigen	(-)
Occult blood	(+)	CRP	(+)
8. Blood biochemistry		RA	(-)
Serum protein	7.6 g/dl	ASLO	50×
A/G	0.92	LE cell	(-)
α1 Globulin	4%	IgA	240 mg/dl
α2 Globulin	9	IgM	90 mg/dl
β Globulin	14	IgG	1,800 mg/dl
γ Globulin	25		

Microfilariae in the peripheral blood negative. FPT reaction (filarial intradermal reaction) positive. VBS reaction (intradermal reaction for paragonimiasis) negative. Slight anemia is present and eosinophilia is not so clear. Erythrocyte sedimentation rate is 27 mm in one hour (Westergren), which is slightly accelerated. Hypergammaglobulinemia is present as well as slightly high level of serum Ig-G and slightly low level of cholin-esterase. Proteinuria and urinary occult blood are moderately positive.

DISCUSSION

Kagoshima prefecture is on the southernmost part of the Japanese mainland with a favorable mild climate and much rain. Tokunoshima is, in particular, a small island which has an area of about 250 square kilometers located to the south of Amami-honto (main Amami island) which is about 500 kilometers from the Kagoshima mainland. The annual mean temperature there is 21 C and it is over 20 C for eight months from April through November, which is thought to agree with the growth of filarial larvae and mosquitoes.

The result of filarial investigations made in Tokunoshima from prewar period until 1960 is shown in Table 3 which reveals 153 microfilaria positive persons were

TABLE 3 Results of filarial investigations in Tokunoshima

Tokunoshima Island	Localities	No. of persons examined	Mf. positives	Mf. positive rate (%)	Reported by	year
	Toku- noshima	118	47	39.8	Yoshinaga <i>et al.</i>	1913
		216	16	7.4	Sameshima <i>et al.</i>	1954
		251	37	14.7	Sato <i>et al.</i>	1955
		392	43	11.0	Fukushima <i>et al.</i>	1960
	Isen	27	4	14.8	Sato <i>et al.</i>	1956
		41	6	14.6	Sato <i>et al.</i>	1956
		1,045	153	14.6 (%)		

found among 1,045 examined (microfilaria positive rate 14.6%). For eight years from 1962 an anti-filaria policy sponsored jointly by central government and Kagoshima prefecture was taken and great results were realized consequently. The result of a government-sponsored anti-filaria work performed in 1962 by Kagoshima prefecture shows 8,968 microfilaria positive persons were detected out of 135,557 examined (microfilaria positive rate 6.6%). As far as the inhabitants of Tokunoshima were concerned 1,254 microfilaria carriers were detected among 10,642 persons examined as shown in Table 4, which reveals considerably higher microfilaria positive rate of 11.8%. All microfilariae were *Wuchereria bancrofti* species. As described above, Tokunoshima has been heavily infested with filarial worms since prewar period and all the worms detected there belonged to Bancroftian species. In connection with

TABLE 4 Results of filarial investigations (Kagoshima prefecture, 1962)

Localities	No. of persons examined	Mf. positives	Mf. positive rate (%)
Tokunoshima	4,454	662	14.9
Amagi	3,133	246	7.9
Isen	3,055	346	11.3
	10,642	1,254	11.8 (%)

the filarial symptomatic patients in Tokunoshima Sameshima *et al.* (1954) reported that they had found eighty patients including twelve hydrocele and twelve elephantiasis ones respectively. However, thanks possibly to filarial mass treatment carried out since 1962 microfilaria positive rate has decreased lately and in 1969 it decreased as low as 0.8% (40 microfilaria positives out of 4,610 examined) in Tokunoshima. With the decrease of microfilaria positive rate number of filarial symptomatic patients has also fallen off remarkably.

According to reports in the past, Blanchard (1900) reported a case of 224 pound scrotal elephantiasis and Pelletier (1912) a 100 kg one. The largest scrotal elephantiasis case ever reported in Japan is one found by Akaboshi, which weighed 63 kg and was 185 cm in transverse and 183 cm in longitudinal circumference. The case of elephantiasis presented in this paper is rather smaller in size when compared with those 'giant' scrotal elephantiasis cases reported in the past. However, in addition to scrotal elephantiasis, the case is accompanied by penile elephantiasis, which is thought to be very rare. The patient has been of mental weakness that it was very difficult to take medical history directly from him and we were obliged to depend on his families and neighbors for the most medical informations on the patient.

A case of huge penile and scrotal elephantiasis accompanied by that of the left lower extremity is presented in this paper.

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巨大な陰茎，陰嚢象皮病の1例

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フィラリア症は，フィラリア性熱発作，リンパ管炎，乳び尿，陰嚢水腫，象皮病等の多彩な症状を呈する疾患である。鹿児島県はパンクロフト糸状虫症の濃厚な流行地であり，特に奄美大島地区は高度な浸淫がみられ，1962年のフィラリア検診の結果でも Mf 陽性率は11.8%で，かつては多くの典型的な象皮病患者がかなりみられた。然しながら最近では典型的な象皮病をみる機会は減少しつつあり，著者が行った鹿児島坊津町清原地区における，1954年から1965年にいたる11年間のフィラリア有症者の追跡調査の結果をみると，陰嚢水腫3例，乳び尿7例の新発生をみているが，象皮病の新発生は1例もなかった。すなわち乳び尿等は新発生があるが象皮病患者は半減していた。最近我々は，巨大な陰茎，陰嚢象皮病患者を経験したので報告する。症例は鹿児島県大島郡徳之島在住の44才の男性である。生来の白痴で IQ は測定不能であった。生後13才迄は蚊の多い山間の家に住んでいた。14才の頃から月に1～2回熱発作があり，30才迄続いた。17才頃から陰茎が異常に大きくなりだして34才の時には膝位の長さになった。34才の時保健所の検診でマイクロフィラリア陽性といわれ治療を受けた事がある。当科初診時，陰茎長は恥骨上縁から 58 cm，陰嚢周は 76.8 cm，冠状溝周 43.5 cm，亀頭周 50.5 cm であった。陰嚢，陰茎を合わせた重量は 18.5 kg（患者体重 52.6 kg）もあった。外尿道口は判然としなかったが，亀頭先端に不規則にみられる溝から尿が滲みだしていた。各種の検査を行ったが EPT 皮内反応が陽性であり%赤沈値中等度促進，軽度の貧血，尿蛋白（+），CRP（+）の他には特に異常所見はみられなかった。