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## Some Problems on Radiotherapy of Cutaneous Hemangiomas

By

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## 皮膚血管腫の放射線治療について

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過去に放射線治療を受けた再来患者を含め、最近5年間に来院した皮膚血管腫 320例 (373個) の診療経験によると、Hemangioma cavernosum の自然治癒率はきわめて高く、法則性をもっている。しかし他方では、皮膚血管腫に対する放射線

治療後の障害はほとんど見られず、発癌もなかった。従つてその治療方針は症例毎に取捨選択すべきで、後障害を考慮しながら適量の放射線治療を行うことは、多くの不幸な子供達に利益をもたらすものと考ええる。

Recently, some discrepancies may be seen on the treatment policy of cutaneous hemangiomas<sup>3)</sup>. In our clinic, a large number of cases have been treated by external or interstitial Ra, external <sup>32</sup>P & Be-window tube soft X-ray (HVD: 3—13 mm) since 1951. Among them, 320 cases have been consulted in 1963—1967. Clinical experience with these cases is presented as follows.

## Clinical Classification and Incidence

Table 1. ♂ : ♀ = 108 : 207. Right : Median : Left = 140 : 51 : 167. Head & neck : Upper extremity and trunk : Lower extremity = 60 : 16 : 5 (Portwine stain), 146 : 98 : 18 (Hemangioma cavernosum). Each incidence decreases from cranial to caudalwards. Though right and left almost symmetrically distributed, mixed type—lymphangioma may prefer left, 55 : 24. On the focal multiplicity, single : double : triple or more = 273 : 43 : 4, majority of the multiple hemangioma belongs to multiple hemangioma cavernosum. Wide area is involved in the large number of portwine stain. Portwine stain, telangiectasia and cavernous hemangioma in a narrow sense are the three elemental types to classify the cutaneous hemangiomas. These types are either compounded or transferred each other during the growing or involuting process, and mixed or strawberry type is the perfected type and occupies a large majority of the cases.

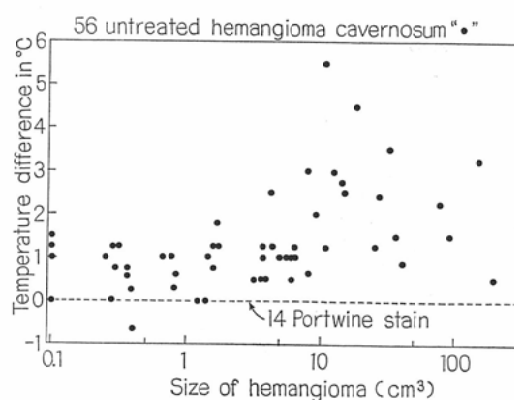
## Angio-somato-dystrophy

Many hemangiomas complicate with local overgrowth, such as hypertrichosis, hyperhidrosis, hypertrophy of bone and soft tissue etc. The larger the hemangioma is, the more prominent these phenomena are, and the type of hemangioma may not be confined to either portwine stain<sup>6)</sup> (Dr. Proppe) or varicosity.

Table 1. Classification and incidence of 373 cutaneous hemangiomas.

		No. of hemangioma	No. of large hemangioma
Hemangioma simplex	1. Portwine stain	86	55
Hemangioma cavernosum	2. Strawberry mark	139	15
	3. Cavernous hemangioma (localized cavernous type)	8	2
	4. Teleangiectasia	8	—
	5. Mixed hemangioma (1,2,3,4)	109	21
	6. Lymph-hemangioma (diffuse cavernous type)	11	8
	7. Mucosal hemangioma	9	—
	8. Specific type (Granuloma pyogenicum etc.)	3	1
Large case: Over $5 \times 5 \text{ cm}^2$		373	102

Fig. 1. Thermometry of the cutaneous hemangiomas. Temperature difference versus size of the hemangioma.



Klippel-Trenaunay-Parkes-Weber syndrome is the extreme manifestation of these phenomena. Considering the possible cases of undergrowth, the expression of "Angio-somato-dystrophy" including many local tissues, may be better than that of "Angioosteohypertrophy"<sup>22)</sup> (Dr. Tobler), of which concept is restricted to the bony structure alone. In the aged lymphangioma or mixed type, local X-ray film often reveals calcified deposits.

#### Thermometry of the cutaneous hemangiomas

With a thermistor thermometer, temperature difference between the surface of hemangioma and its surrounding skin surface was measured. Fig. 1. The surface of hemangioma cavernosum is warmer than the surroundings, nearly proportional to its size, and the temperature difference reaches max.  $5.5^{\circ}\text{C}$ . Progressive hemangioma is somewhat warmer than the regressive case, and the older the children, the temperature difference may be the less. Strawberry type is the warmest, cavernous type is in the next and

Fig. 2. Difference of the age distribution between portwine stain and hemangioma cavernosum at the first consultation.

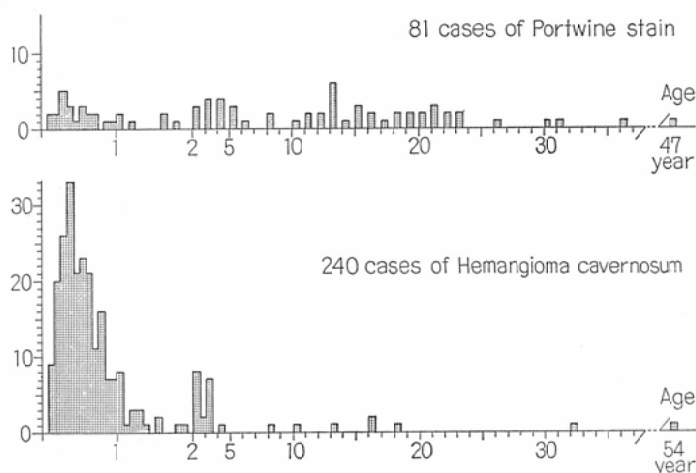
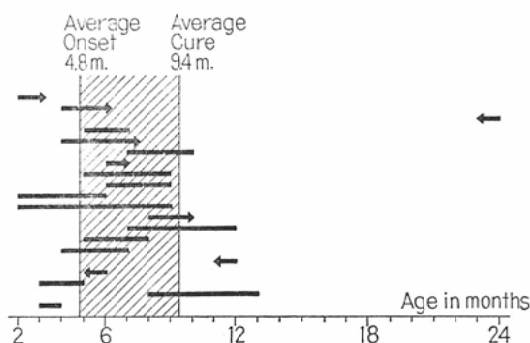


Fig. 3. Observation of 20 spontaneous necrosis of the nonirradiated hemangioma cavernosum.



portwine stain shows no difference. In case of wide strawberry mark, periphery is warmer than the central region.

#### Course observation in the untreated cases

Fig. 2. shows the age distribution of our case material at the first consultation. Distribution curve of portwine stain is quite different from that of hemangioma cavernosum, the former appears to prefer both adults and elder children. This means that a large majority of hemangioma cavernosum heals satisfactorily either spontaneously or by irradiation, but portwine stain is not the case. In regard to the age of onset, almost all portwine stains are congenital, but the onset of hemangioma cavernosum distributes as if the composite of both congenital and acquired, of which median onset age is the 8th day from birth. Nearly all hemangioma cavernosum grow at their early stage, but the growth is interrupted subsequently and finally the regression is replaced slowly. Standstill of the growth appears at the average of the third month from birth and regressive sign appears at the seventh month from birth. Spontaneous necrosis, being the extreme manifestation of this regression, takes place mostly between the fifth and the ninth month. Fig.

Fig. 4. Comparison of the regression rate of hemangioma cavernosum between the watched case group and the irradiated group.

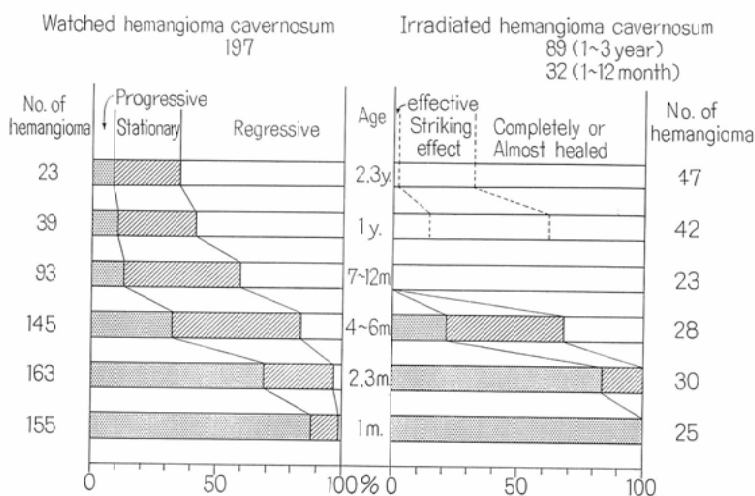


Fig. 5. A persisted adult case of hemangioma cavernosum on the face, biopsied at the Departm. Dermat. Osaka Univ. Med. School. 54 year old male.

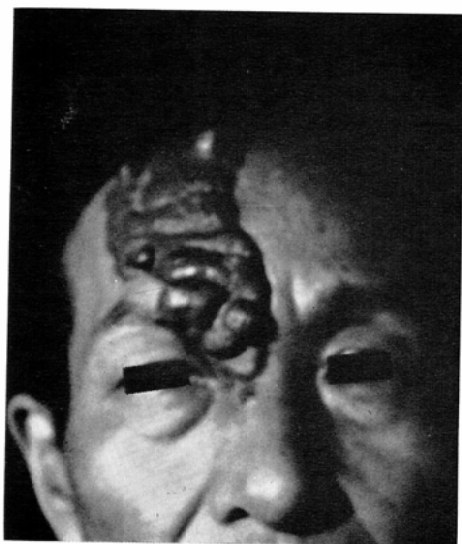
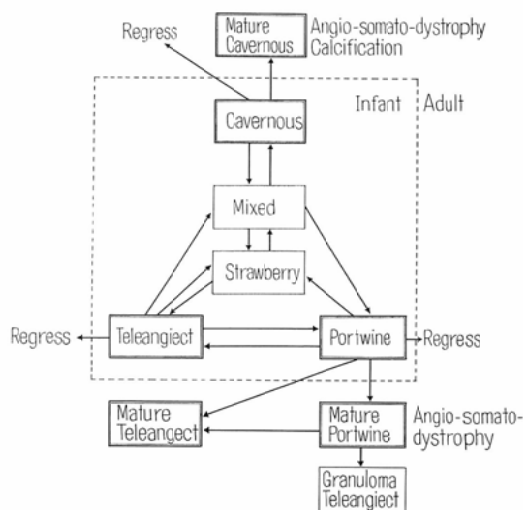


Fig. 6. Diagrammatic illustration of the classification and the spontaneous or post-irradiative course of cutaneous hemangiomas.



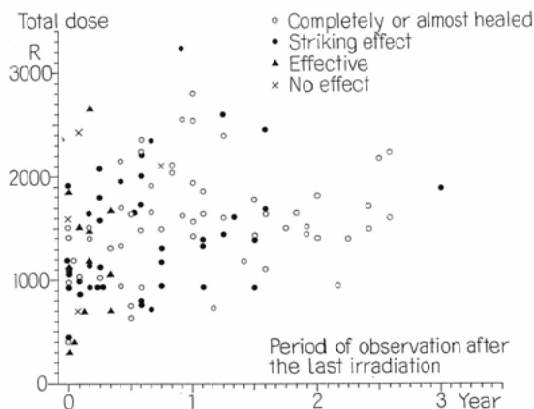
3. By statistical observation of the nature of hemangioma cavernosum at the given age, frequency of the spontaneous involution was estimated. Fig. 4. Left. As either completely or almost healed cases may not necessarily be consulted, the numerical values mean the minimal spontaneous regression rate. The rate reaches 65% at 2—3 years of age, if the type is limited to the pure strawberry, the rate reaches 77% at this age. The true regression rate may be estimated far higher than these rates, and probably be nearly 100%, observing for longer period. Fig. 5. illustrates one persisted adult case of hemangioma

cavernosum of the face. Onset age of the disease does not correlate with the onset age of regressive sign. We have experienced several suddenly and rapidly involuting cases referred to an unrelated etiologic factor, such as the complication of measles, pneumonia or enteritis etc. There were neither malignant nor hospitalized cases. So-called mutilation, such as the perforation of auricle or the deformity of nose etc., is frequently encountered in the early progressive stage, so that hemangioma cavernosum in this stage has "semimalignant" property as pointed out by Dr. Bredt<sup>1)</sup>. Spontaneous regression rate of the small and faint portwine stain is fairly high, but cases of larger and deeper color regress scarcely. Fig. 6. shows diagrammatic illustration of either spontaneous or post-irradiation course of cutaneous hemangiomas, presumed by our experience.

### Therapeutic effect of radiation

When the superficial X-ray therapy of 100—300 R/3—6W is continued, spontaneous growth of the hemangioma cavernosum is suppressed, stationary hemangiomas turn into regressive and involution of the hemangioma is accelerated. Observing 42 progressive or stationary hemangioma cavernosum before irradiation, all cases become regressive after the average three months from the first irradiation. Complete cure or striking effect is achieved by an early radiation therapy within the age of 2—3 for all cases. Fig. 4. Right. Even a small total dose of 300 R is sometimes very effective. The more the single dose is,

Fig. 7. Relation between the effect of radiotherapy, the total tumor dose and the period of observation after the last irradiation of 113 hemangioma cavernosum.



the more rapid involution may appear, but according to Fig. 7. any clear correlation is not found between the total tumor dose and the effect of radiation. Therapeutic effect correlates with the period of observation after radiotherapy rather than with the total dose, and nearly all cases of hemangioma cavernosum disappear either completely or on the whole after 1.5 year from the last irradiation. This fact has been already pointed out by Dr. Schondorf,<sup>7)</sup> and our opinion on radiotherapy of hemangioma cavernosum is the followings: radiation acts as if antibiotics to the infections disease, suppresses the pathologic process and self control mechanism of the organism is finally participated. Both spontaneous or post-irradiative regression appears to take frequently the reverse process to its initial growth. Radiation is also considerably effective for cavernous hemangioma of the mature type, except for adult cases. All cases of portwine

stain or lymphangioma of the infants are not radioresistant and not a few children with these types may be benefited by the reasonable dose of radiation.

### Radiation sequelae

Among re-consulted 38 patients, whose cutaneous hemangiomas were irradiated many years ago, 20% of them have consulted for fear of the radiation hazard. But remaining 80% have consulted with the request of re-irradiation expecting better cosmetic result, and the large majority of them were portwine stain. Table 2. On the abnormality of the skin, bone and cartilage, teeth, breast or thyroid gland, frequently argued on radiation damage, both the early invasive stage of the hemangioma itself and the possible angio-somato-dystrophy syndrome, above mentioned, should be considered, so that the statistical control with nonirradiated similar cases is required for the definite diagnosis of late radiation damage.

#### 1) Skin and subcutaneous tissue

Hyperpigmentation of the skin after superficial X-ray therapy disappears three years later, but as the slight hyper- or hypo-pigmentation is quite elusive for the repeated long-term observation, definite diagnosis is difficult. Hypofunction of sebaceous or sweat-gland of the local skin is also of temporary nature. Though either slight atrophy of the skin or teleangiectasia may be observed in the majority of our cases, differentiation from the spontaneously involuting process is difficult, as argued by Dr. Luger<sup>5)</sup> or Dr. Klostermann<sup>4)</sup>. As a rule, it may be impossible clinically to detect the effect of past irradiation, several years after the superficial X-ray therapy. Chronic dermatitis may probably be the most significant late complication of the cutaneous hemangiomas.

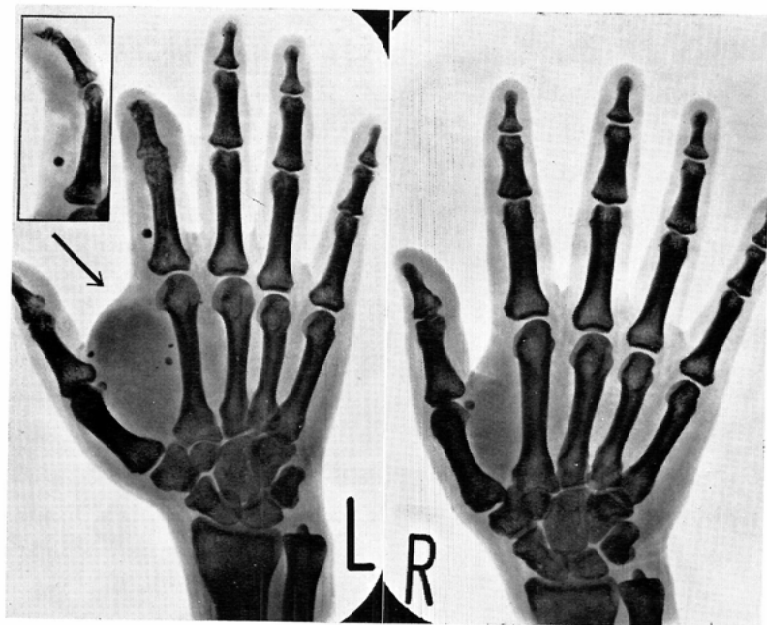
#### 2) Bones, Joints and Teeth

Routine X-ray examination of the local irradiated tissues disclosed bone abnormality of the hand in 2 cases, Ra-needled lymph-hemangioma and with <sup>32</sup>P externally treated portwine stain. Shortening, slight curving and osteoporosis were observed in the former case, and slight deformity was found out in

Table 2. Analysis of the reconsulted cases many years after the radiotherapy of cutaneous hemangiomas.

Chief complaint	No. of cases	Techniques of past irradiation	Interval (year)	Diagnosis at re-consultation
Request for re-irradiation	29	Ra, <sup>32</sup> P, X-ray <sup>c</sup>	Average 4	
Local pain	2	Ra-needle, <sup>32</sup> P	8, 7	Radiogram of bone & joint: abnormal 1
Local eczematous eruptions with bleeding tumor	1	Ra, <sup>32</sup> P	9	Chronic dermatitis Suspicious of Granuloma pyogenicum
Marked local scarring	1	<sup>90</sup> Sr ? Ra	10	Slight hemihypertrophy (diseased side)
Local tumor with bleeding	1	<sup>32</sup> P	6	Granuloma pyogenicum
Epilepsy	1	<sup>32</sup> P	10	Sturge-Weber syndrome
Local bleeding	1	<sup>32</sup> P	3	Hematologic exam. normal
Weakness	1	Ra, X-ray	2	Hematologic exam. normal
Phobia for breast damage	1	Ra-needle	12	Symmetric develop
	38			

Fig. 8. Radiogram of both hands of a 19 year old female with lymph-hemangioma on the left, which was treated by Ra-needle 8 years ago, and thereafter operative procedure was repeated.



the latter. Fig. (8). It may, however, be difficult to judge simply that these abnormalities have a direct relationship to the past radiotherapy. Therefore, growth of the local bones before or after radiotherapy was compared radiographically to the opposite normal side on 34 cases of cutaneous hemangioma of the extremities. As a result, slight tendency to the more rapid growth of diseased side than normal side was observed, having no relation to radiotherapy. The older the children are, the more prominent the difference of bone-growth of both sides appears. Observations on either the appearance of ossification center or the asymmetry of the skull could not find out any damage to bone-growth. No abnormality of the dentition was observed, and several cases with earlier dentition of the diseased side was observed after the total dose of 3000 R

### 3) Eye etc.

As we have irradiated with Pb or Au protective shells<sup>8)</sup>, damage to the deeper portion than cornea might not be considered. Permanent hyperemia of the conjunctiva was observed in some cases of aged portwine stain treated by either  $^{32}\text{P}$  or Ra. Except them, late damage of the conjunctiva was not observed. Abnormality in the blood examination, damage to the brain or local malignoma was not experienced.

### Summary

Clinical experience with 373 cutaneous hemangiomas informs that though spontaneous involution of the hemangioma cavernosum is quite frequent, one encounters both the real radiation damage and radiation induced malignoma seldom or never. Therefore, the treatment policy of cutaneous hemangioma depends upon circumstances, and adequate dose of radiation considering the late damage may be of definite value for the many unfortunate children.



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