

INVESTIGATIONAL STUDIES IN SOME CONGENITAL AND ACQUIRED DEFECTS OF THE HAIR IN CHILDREN*

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In the perplexing field of the study of the changes of hair, often many things are more apparent than real. One longs for the infinite patience and detailed experience of Pinkus (1, 2). The clinical reviews of Sabouraud (3), Galewsky (4), Savill (5) and McCarthy (6) are well known. Up until recent years it has been the anatomists, the anthropologists, the endocrinologists, those interested in forensic medicine, and those working in the fibers of animals who have made the significant advances in the critical study of hair. The return of the dermatologists to basic work in the study of hair has been led by Rothman and his co-workers (7, 8). The resurgence of the spread of tinea capitis has emphasized to the dermatologist the necessity for increasing knowledge of the hair.

The difficulty of a critical examination of change in structure of hair as relates, for example, to growth, size, texture, pigment, and the like, has been mentioned. "Those who have tried to judge slight color change in the skin or hair realize how easily one can be deceived" (9). The small degree of response, if any, the prejudice of the patient and the observer, the variability of the individual, and the limitation of the instruments of precision are but some of the factors which serve to confuse the observations. Because of the "greater range of reaction of children" and the apparent greater ease of their hair to change, the general disinterest of young children in their hair, the availability of clinical material, it was decided to initiate the investigational procedures in a group of children.

METHODS OF STUDY OF HAIR

When it is possible, the detailed examination of the hair should proceed in an orderly fashion. Briefly, the gross examination of the hair in the scalp should include:

1. Examination in daylight or incandescent light.
 - A. General appearance
 - B. Anthroposcopic observations
 1. straight
 2. low wave
 3. medium wave
 4. deep wave
 5. curly
 6. frizzly or kinky
 7. coiled or spiral tufts
 8. pepper corn

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C. Color

1. natural or artificial
2. shade
3. distribution

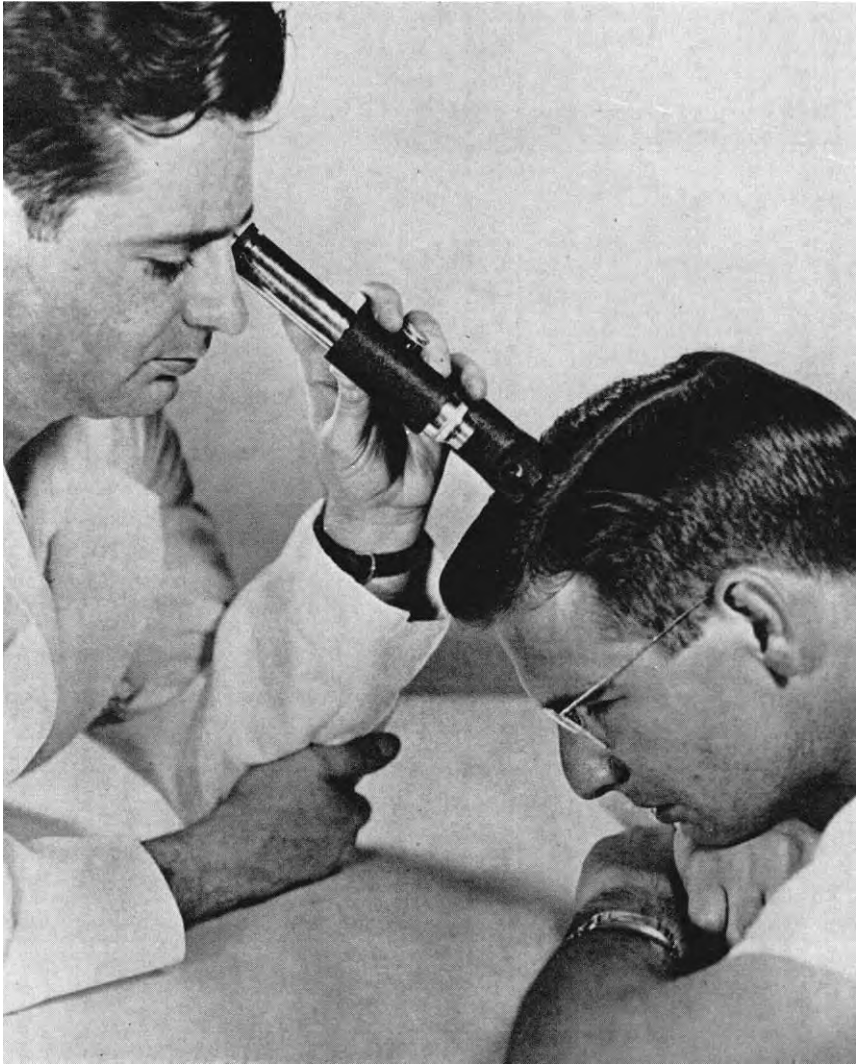


FIG. 1. THE USE OF THE SKIN MICROSCOPE AS AN AID IN THE FLEXIBLE MICROSCOPIC EXAMINATION OF THE HAIR IN SITU

- D. Hair length—measurement if desired
 - E. Correlation of changes in hair of scalp with hair elsewhere on body
 - F. Correlation of changes in hair with possible changes in skin, nails, etc.
 - G. Hair count in a certain area
2. Examination in ultra-violet light through a Wood's Filter

3. Photography—unless the changes are marked, gross photography including koda-chrome photography, is of limited value.

The examination of the hair in the scalp may be continued by means of skin microscopes (10, 11). These may include the flexible portable scopes, 20 \times , 40 \times or 50 \times , or the less flexible stereoscopic binocular up to 108 \times . The simple pocket microscopes, especially the Optiker Peschke Nurnberg type (40 \times -60 \times), may also be used. A self-contained source of illumination would make these small instruments even more practical. The advantages of direct cutaneous microscopy include:

1. Examination of the hair in situ.
2. First opportunity to examine cortex and medulla.
3. Detailed examination of the medulla with transmitted, vertical or oblique illumination of the hair.

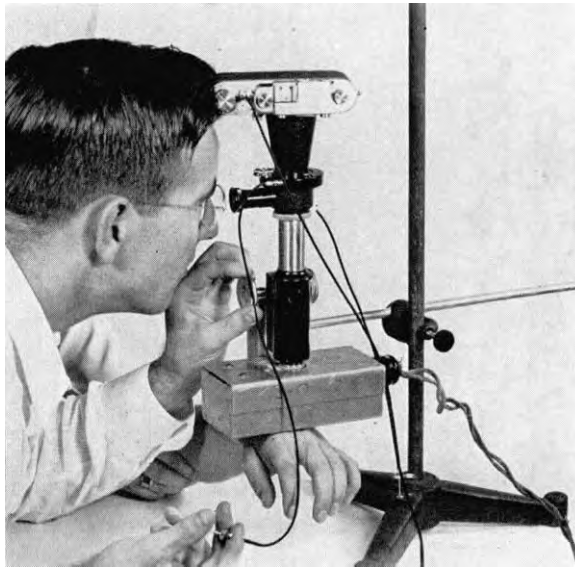


FIG. 2. CUTANEOUS PHOTOMICROGRAPHIC APPARATUS OF SIEBENTRITT FOR PHOTOMICROGRAPHY OF THE HAIR AND FOLLICLES IN SITU

4. Distribution and character of the pigment granules.
 5. The measurement of the diameters at various positions along the hair shaft.
 6. The detailed examination of the hair tip.
 7. The early detection of new growth of hair.
 8. Detailed and accurate hair count on a small area.
 9. Character of the mouth and position of the follicle.
 10. Study of the associated surface of the scalp.
 11. Capillaroscopy studies of the skin of scalp if desired.
 12. Opportunity for examination of hair and scalp microscopically in ultra-violet light through a Wood's Filter.
 13. Adaptability for a portable cutaneous photomicrography apparatus such as that of Siebentritt (12).
 14. Adaptability as a portable polarizing microscope.
- The next step in the examination is the removal of the hair from the scalp. In our preliminary studies we have done this in an uncontrolled fashion but this may be accomplished

somewhat critically by the following technics. In general the pulling-out weight required varies from 10 to 40 grams.

1. pinch clamp and hook of Basler (13)
2. light wire spring scale of Au and Bogen (14)
3. steel band spring technic of Whitaker (15)
4. pilometer of Copley (16)

After the hair is pulled out the character of the attached hair bulb should be noted. A "good" hair is evident, for example, by a "sukkulenten Wurzelscheiden" of Pinkus. The detailed examination of this may be aided by the skin microscope.

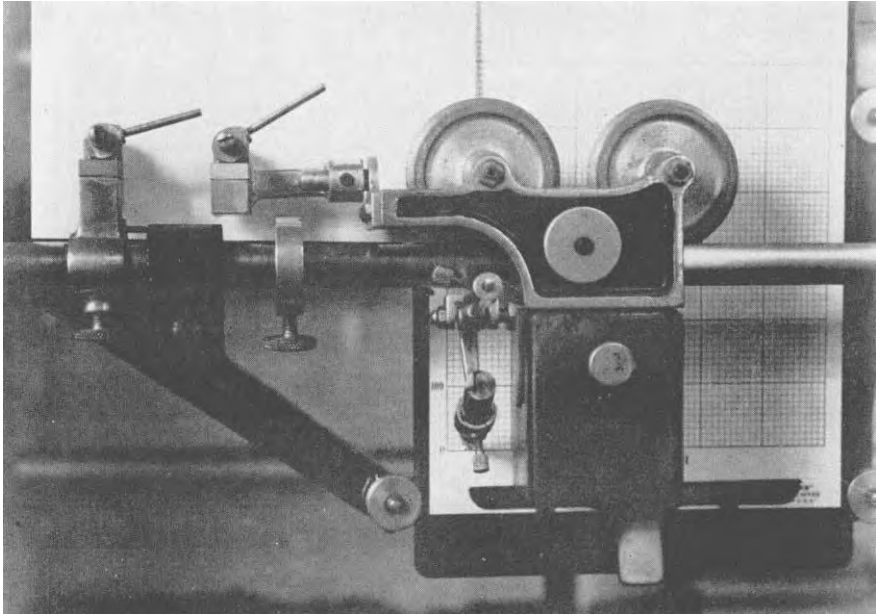


FIG. 3. THE SCOTT FIBER TESTING APPARATUS AN EXAMPLE OF THE INCLINED PLANE TENSILOGRAPH

A constant temperature room is necessary. Speed of reaction fixed in this type.

Now, the physical properties of the hair can be examined. These include elasticity, extension, bending, tearing and compression (17). Elasticity, extension and tearing are the ones more commonly studied. In our preliminary work, we have considered only extension and tearing (tensile strength). In this field many controls appear necessary, for such factors as length, diameter, speed of reaction, hydration, etc. may affect the result. We agree with Savill (5) that X-ray diffraction studies provide the most accurate means of studying elasticity of the hair. For the study of extension and tearing, among others the following technics may be used.

1. inclined plane tensiograph (18)
2. balance technic of Basler (17)
3. Salter Balance Technic of Leftwich (19)
4. pilometer of Copley (16)

The surface of the cuticle may then be studied by imprint technics before a permanent mounting of the hair is made. These technics include:

1. Moritz nail polish technic (20). Colored nail polish appears to us to be superior to films of colorless nail polish.

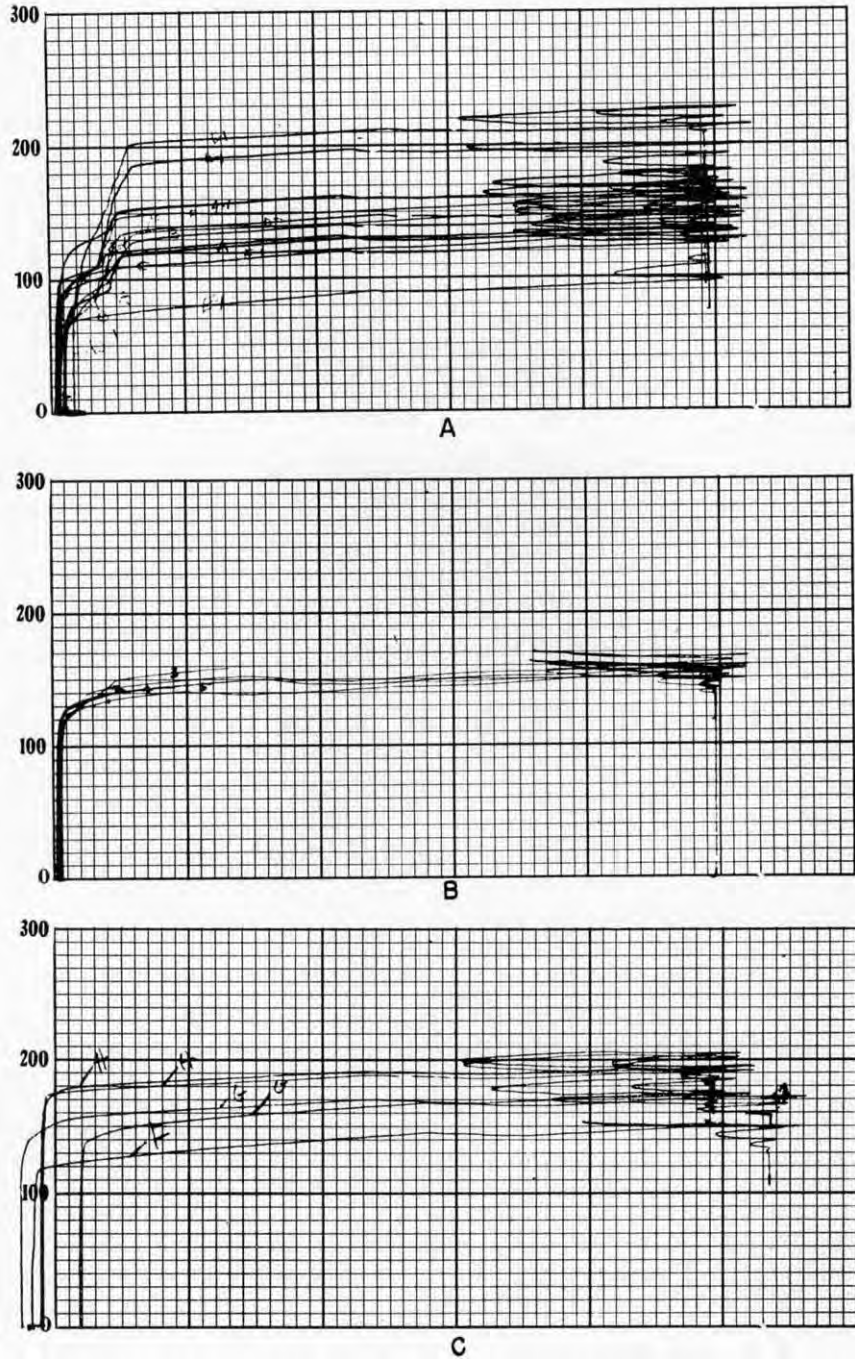


FIG. 4. ILLUSTRATING THE TYPES OF CURVES OBTAINED BY THE INCLINED PLANE TENSILOGRAPH

A. Normal hair. B. Favus hair showing no extension. C. Hair of the idiopathic (deficiency) group.

2. Plastic sheet pressure technic of Hardy and Plitt (21). Instead of the stripping away of the hair from the film of nail polish (Moritz technic), sheets of plastic are forced over and under the hair and imprints are made. Through the efforts of Kooyman (22) we have observed an excellent example of this technic.

The hair may then be prepared for permanent mounting. The hair may be imbedded directly, as we have done with the use of Permoult (23), or the hair may be subjected to action of water, alcohol, ether, alkalis, or acid. The agent used depends upon the details one wishes to observe such as cells, pigment, "air vesicles" etc. The picture of the unstained hair may be recorded in several ways.



FIG. 5. ILLUSTRATING THE SPARSENESS, FINENESS AND DEPIGMENTATION OF THE HAIR OF A CHILD OF THE IDIOPATHIC (DEFICIENCY) GROUP

1. Technic of Maturra as modified by Pinkus'(1) for the projection of detailed measurements on graph paper.
2. Photomicrography with measurements in scale—Photomicrography is difficult at high powers because of cylindrical shape of hair.

For detailed study of the basic structure of the hair prepared microscopic sections are necessary. The difficulty of securing good cross sections of hair is well known. Recently two relatively simple technics have been proposed for such sections.

1. Hardy cross section apparatus (24)
2. Adhesion technic of Garn (25)

Under some circumstances, longitudinal sections of the hair may be desired. Perhaps phase microscopy will be of value for hair sections; it was not of value for unsectioned hair.

Selective staining of the hair may be accomplished, especially with the trichrome tech-

tics. The dopa reaction may be used for the study of pigment. The details of the histopathology may be obtained from standard tests and especially from the work of Trotter (26) who has studied the hair in its relationship to dermatological problems.

The anatomical and simple biophysical studies of the hair are amplified by detailed study of keratin by biochemical technics and the important X-ray diffraction studies, especially as adapted by Astbury (27 and Harris). The hair may also be examined in addition to metabolic products for such foreign substances as dyes, heavy metals, etc.

THE GENERAL GROUPS STUDIED

The cases in children studied have been divided into the following general clinical groups.

1. Congenital abnormalities

1. Hypotrichosis

A. Associated with congenital ectodermal defect—2, only 1 observed over a prolonged period of time.

B. Idiopathic sparse thin hair type—1 (observed only 2 months) "true hypotrichosis"

2. Congenital familial premature canities—1 (onset at age of 5)

3. Monilethrix—1

4. Pili torti—1, not observed over a prolonged period of time

2. Acquired abnormalities

1. Central American avitaminotic group—2

2. Association with chronic intestinal parasitism (oxyuris)—2

3. "Systemic Idiopathic" (possible deficiency)—4

4. Fungous infection

A. *Microsporon Audouini*

1. with topical chemotherapy—6

2. post epilation—11

B. Endemic favus group—under study at present

5. Premature unilateral calvities frontalis—1 (onset at age of 9 years)

No special efforts were made to study the growth characteristics of hair in the new born. In general, save for congenital abnormalities, recognizable hair disturbances in young children were uncommon. It is certain that we have missed many slight changes. At the Children's Hospital, children with chronic illnesses were observed from time to time in relationship to their scalp and hair. Little gross changes could be made out.

We agree with Stevenson (28) that congenital syphilis appears to be able to produce temporary hair loss in children more easily than the other childhood diseases, including the infectious exanthemata. Many of the cases of so-called "idiopathic systemic" group were recognized by an observant (as opposed to a prejudiced) mother. When our experience is greater and our technic of examination more precise, then perhaps we may find more cases. It is curious how much the disturbances of keratinization of the skin, such as severe ichthyosis, vitamin A deficiencies, etc., are so little reflected in the disturbances of the scalp and hair.

The severe and varied avitaminoses associated with hair changes in white and mestizo children in the American tropics have been reported before (29). Additional material has been sent to us from our associates at San Juan de Dios Hospital in San José, Costa Rica under the direction of Peña Chavarría. As yet we have not had opportunity to study scalp biopsies in these cases. Additional proof that this syndrome is a definite one in the tropics

are the recent reports by Hughes (30) in negro children and Nichols (31) in Malays and Tamils. Karefa-Smart (32) has also mentioned the peculiar yellowing and blonding and straightening of hair in children with severe untreated hookworm infestation in Sierra Leone, West Africa. Hairs from these cases have not been examined as yet by us. This so-called "grizzling" reaction of the hair of children in the tropics still remains uncommon, even with the severe nutritional cutaneous reactions found, such as even the bullous, depigmented, exfoliative and gangrenous cutaneous types. Rao (33), from his vast experiences with nutritional disturbances in India has claimed that he has never seen such hair changes. Moreover, as we reported in 1946, no such hair changes have been observed in the nutritional disturbances of children in the United States. Perhaps there is a similarity to some of the changes of a slight nature which we have observed such as in two cases of severe pinworm infestation and in four cases where we could ferret out a possible "intestinal factor" (diarrhea, prolonged mineral oil medication, sulfonamide therapy, etc.). In the cases in Costa Rica, depigmentation would recur when adequate therapy (dietary chiefly) again was not possible. It was difficult to determine in some of our cases whether the effects of initial therapy were permanent. In two cases for example, substitution of placebo therapy for calcium pantothenate caused a recurrence of hair loss but no evident change in color. Perhaps as in animals, the bacteria flora in children is important in the development of achromotrichia and as Nichols (31) states there may be then a "deficient nutrition of nerves." From a clinical aspect, sun depigmentation (more spotty, no changes in diameter, no alopecia) and normal darkening effect of hair physiologic tricolorosis, (not reversible, no change in diameter) appeared to be conditions to be ruled out in a consideration of "avitaminotic" achromotrichia of children.

Cases of fungus infection of the hair were included to attempt to determine, if possible, the general aspects of the relationship of nutrition to the clinical picture. This phase is being studied in the endemic favus in certain areas of Kentucky. Cases of *Microsporon audouini* infection were selected to determine whether feeding experiments could influence the topical therapy or the return of hair after X-ray epilation.

EXPERIMENTAL OBSERVATIONS

All cases were recorded on detailed hair charts. All patients had microscopy of the scalp and hair by the skin microscope, frequently under ultraviolet light through a Wood's filter. These preliminary observations often revealed a greater degree of change than was visible with the eyes. The hairs were then cut off or pulled out and mounted in Permount. These mounts enabled the hair to be studied in still greater detail, enabled more accurate measurements of diameter than could be done on the scalp. Permanent records of the hair of each case were then available. Hair counts were made with the 0.5 cm² rubber stamp. In general, there was so much variation that a general impression of sparseness was as accurate as the count. Our general average was low, 20-25 per 0.5 cm². Because the margin of error may be so large with such a small stamp, a larger field will have to be chosen. Cuticular patterns were done, chiefly with the Moritz (20) technic, and showed no recognizable changes save perhaps in severe fungous infected hairs. The furrows in the film showed often a spotty fluorescence after the infected hair was pulled out of the nail polish film on the slide. Accurate patterns with examination under high magnification may enable us to determine variations more easily. It is hoped also that shortly X-ray diffraction studies will be available.

This table may show changes as the child grows and the "lack" of elasticity of

fungous infected hair and perhaps diminished elasticity of the "deficiency groups". Whether the tensile strength is still maintained by the medullary structures is not evident in this small series. Not enough material was available from the follow-up of the "Idiopathic" (Deficiency) group to determine whether clinical improvement is associated with change in the "Tensigram" picture. With careful control conditions (including variations in speed) of the hairs to be examined, and uniformity of result with this apparatus, it does appear possible to detect minor variations of the properties of elasticity, extension and tensile strength.

We have not had enough experience with the prepared section technics to determine their practical value in these cases. If good sections could be prepared routinely, it is possible that small differences in anatomical detail could be ascertained.

TABLE I

Results with the use of the inclined plane tensiograph in hair of children (hair length 2.5 cms.)

DISEASE	AGE GROUP	NUMBER	AVER- AGE DIAM- ETER HAIR	EXTEN- SION	TENSILE STRENGTH (TEARING)
			mm.	mm.	gms.
Normal.....	1 mo.-5 yrs.	24	0.0396	6.4	97.6
Normal.....	5-15 yrs.	11	0.061	7.7	124
Sun Depigmentation (portion).....	5-15 yrs.	16	0.084	10.0	172
Tinea Capitis.....	5-15 yrs.	8	0.070	0.0	150
"Idiopathic Deficiency" Group.....	5-15 yrs.	6	0.070	6.7	103
Alopecia Areata.....	5 yrs.	1	0.087	10.0	140
Hypotrichosis (Cong. ectodermal defect).....	10 yrs.	1	0.112	5.0	140
Rheumatic Fever.....	12 yrs.	1	0.037	10.0	150

The feeding experiments in the children with fungous infection of the hair revealed nothing definite. It was not possible to control the experiments in detail. Calcium pantothenate (150 mgs), panthenol (100 mgs), with or without B complex capsules did not appear to enhance the value of any form of local therapy and it was not possible to tell whether or not the return of hair after X-ray epilation was accelerated. The severity of endemic favus in some sections of Kentucky is a better source of material because of the resistance of the disease to therapy and the definite suspicion of associated nutritional factors. The results in this study will be reported at a later date.

Many other cases in children of seborrheic dermatitis, ichthyosis, etc., were discarded because the period of observation was not long enough or not controlled well. Fifteen cases of alopecia areata were treated with either calcium pantothenate (150 mgs) or panthenol (100 mgs) but these therapies did not appear to influence the result. Only three cases of post-infectious alopecia were treated with calcium pantothenate (150 mgs) and again it was not possible to say if this therapy had any influence on the return of hair.

Seventy adult patients, at present, have also been studied with calcium pantothenate, panthenol and d-biotin therapy of a variety of scalp and hair

TABLE 2

Clinical results—Summary of results of systemic therapy as effecting color and growth of the hair

CASE	PERIOD OF OBSERVATION	THERAPIES (SUMMARY)	RESULTS		REMARKS
			Hair color	Hair growth	
Hypotrichosis with congenital ectodermal defect	18 months	Calcium pantothenate (150 mg.) B Complex Capsules (2 daily) Biotin (0.25 mg.—2 daily) Vitamin A (100,000—2 daily)	No change	?	No change in skin or nails
Monilethrix	2 years	Vitamin A (50,000—2 daily) Calcium pantothenate (150 mg.) Biotin (0.25 mg.—2 daily) B complex capsules (2 daily)	No change	No change	
Congenital familial premature canities	8 months	B complex (2 daily) Calcium pantothenate (200 mg. daily)	No change	No change	
Premature unilateral calvities frontalis	1 month	Calcium pantothenate (200 mg. daily)	No change	No change	
Idiopathic alopecia with					
1. pinworm infestation	6 months	Calcium pantothenate (150 mg.) B complex capsules (2 daily)	No change	Definite improvement in 3 weeks	Infection not controlled
2. pinworm infestation	2 months	Calcium pantothenate (150 mg.) B complex	No change	Improvement, 1 month	
3. feeding problem	10 months	Biotin (0.25—3 daily) Calcium pantothenate (150 mg.)	?	Improved 2 months and continued after therapy stopped	
4. ?	12 months	B complex capsules (2 daily) Calcium pantothenate (200 mg.)	Darkened	Improved	B complex alone would not hold child. Calcium pantothenate alone maintained hair growth.
5. ?	12 months	Calcium pantothenate (100 mg.)	No change	Improved	
6. Intestinal factor Mineral oil and repeated sulfamide therapy)	1 month	Calcium pantothenate (100 mg.)	No change	No change	

conditions, and vitiligo. In all save two cases, (d-biotin plus calcium pantothenate) the results were completely negative. No para-aminobenzoic acid was used in this series. For investigative studies, panthenol has been studied in

buccal lichen planus and massive calcium pantothenate therapy (600–800 mg) in subacute and chronic discoid lupus erythematosus (34). In 1946, ninety-one patients with hair and scalp disorders, vitiligo, acne, seborrhea (35), etc., were treated with 0.50–0.75 mgs. of biotin¹ daily, often over prolonged periods of time. Save for the avitaminotic hair changes in children, no definite and unequivocal results could be observed.

SUMMARY AND DISCUSSION

Even from our preliminary observations, it appears more than coincidence that some children otherwise in "good health" should reveal acquired and reversible changes in hair. The majority of these cases had some intestinal difficulties. Perhaps these cases resemble the more severe manifestations that are seen in children in the tropics. Perhaps also these cases resemble the so-called nutritional achromotrichia of experimental animals. Because of the greater ease of reaction of children, detailed experimentation should be continued in this group and efforts be made to determine the significance and mechanism of these transient pathological changes in hair. Then, we can proceed to the infinitely more complex and much more controversial adult groups (36, 37).

The important question of the endocrine relationship to changes in the hair in children was not examined critically in this preliminary report, for the most part, on the pre-puberty child. Detailed physical examination notes were recorded on the clinical records and clinical endocrine studies, especially basal metabolism readings, were done as indicated. In adults this endocrine phase may be much more important.

It appears that frequently we miss slight changes because we have not been able to adapt precision instruments to clinical practice. Perhaps the experience and patience of older investigators in this field and correlated fields will help to encourage dermatologists to continue to utilize these aids and to secure more accurate ranges and patterns of controls.

CONCLUSIONS

As the preliminary phase of a critical study of hair, some congenital and acquired defects of hair in children were studied. The detailed technic of study of hair was reviewed and many of the current methods were employed in this series. Microscopy with skin microscopes appeared to aid clinical observations but it is hoped that detailed prepared specimens may offer promise for early detection of significant and slight anatomical changes which are often missed. In children, that group of hair defects, change in pigment and change in growth, which appears to offer greatest promise for study and response to therapy is the acquired form associated most often with some intestinal disturbances which appear "to affect" intestinal flora. This group in the United States may be related to a more severe reaction, the nutritional achromotrichia of the children in the tropics. Calcium pantothenate and panthenol in relatively large doses (necessity?) appeared to influence the return of normal hair development only

¹ Biotin furnished by Merck & Co., Inc.

in these children of our entire series of various congenital and acquired disturbances of the hair.

The authors are grateful to Dr. Herman Pinkus for furnishing literature and of material of the work of Dr. Felix Pinkus, and to Mr. Roddy of the Tanners Research Laboratory of the University of Cincinnati for the use of the Scott Fiber Testing Machine.

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DISCUSSION

DR. ZACHARY FELSHER: I was interested in what Dr. Goldman had to say about the green fluorescent substance in tinea infected hair. There has been some controversy about the source of the fluorescence: does it come from the mycelia, from the spores, is it secreted or kept in situ in the fungus itself? Davidson and Gregory were able to extract the green substance very easily by boiling hair in water. The green substance came out in solution, and in filtering the solution was green. I was also able to extract the green substance by putting hair in 2 N cold sodium bromide solution for about 15-30 minutes. On examination under the Wood's light the green substance is present in the solution. It is also interesting to note that the substance seems to be an indicator. It changes color at various pH, being green in acid pH and blue in alkaline pH. In the sodium bromide solution it is just the opposite, blue in acid and green in alkaline solution. We also attempted to get cross sections of hair, but did not have the nice apparatus that Dr. Goldman uses. We got the cross sections by placing the hair in paraffin and cutting in by the ordinary method. On examination under the Wood's light the cross section showed the green substance inside the hair itself and not simply around it.

DR. FRED D. WEIDMAN: Dr. Felsher brought up the matter of histologic technic in the case of very small materials such as hairs. When they are quite dry, they can be imbedded in paraffin without dehydrating, but even here there are difficulties in orienting them. Of course, when they are wet, they must be dehydrated. In either case, orienting can be effected simply and efficiently as follows. A piece of fresh liver, whether from necropsy or butcher shop, is trimmed into a cube about 1 cm. square. An incision or deep pocket is made upon its surface; into it, the hairs or other materials can be adjusted with a needle according to the needs of the worker. The tissue juices are sticky enough to hold them in place. The block of liver is dropped into the fixative and sectioned secundum artem.

I used this method for sectioning scrapings from black hairy tongue and also fragments of finger nails which had to be adjusted into a composite whole. (Weidman, F. D. Affinities between black tongue and trichomycosis. *Arch. Dermat. & Syph.* **18**: 647-665, 1928;

and Robinson, M. and Weidman, F. D. *Dystrophia unguium mediana canaliformis*. Arch. Dermat. & Syph. 57: 325-331, 1948.)

The introduction of some particulate matter like india ink into the pocket would probably assist the technician to locate the significant material when cutting the sections.

DR. LEON GOLDMAN: We had emphasized in the work done that we had done no critical studies in regard to the retention of fluorescent material in the hair shaft print in the film of nail polish on the slide. For some critical studies we would refer you to the work of Dr. Maurice Strauss of New Haven. Certain mechanical factors would suggest that we were also removing some of the cuticular elements in this printing process. The use of the liver block as Dr. Weidman suggests seems ingenious and certainly much more physiological than the use of rubber cements and imbedding material to keep the hair from twisting and turning. Workers in the field of forensic medicine, however, have not had too much success with this technic of insertion of hair into liver tissue.