

MULTIPLE HAIRS (FLEMMING-GIOVANNINI)

REPORT OF TWO CASES OF PILI MULTIGEMINI AND DISCUSSION OF SOME OTHER ANOMALIES OF THE PILARY COMPLEX*

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Of all the adnexa of the skin, the hair has the most elaborately balanced structure. Only minor deviations from the norm are compatible with the formation of a viable hair. Two of the basic rules of hair morphogenesis are these:

1. One papilla forms only one hair at one time; 2. Each hair is a unit, a single thread made up of concentric layers, and does not branch or become subdivided. These rules hold true not only for human hair. From the finest hair of the chinchilla fur to the coarsest vibrissa of the tiger, hair formation follows the same rigid rules.

This presentation deals with aberrant formations of hair which are exceptions to the "one or nothing" rules. In these cases, several incomplete hairs are present in one follicle, and on gross inspection of the skin, a bundle of hairs seems to protrude from the follicular mouth. As there are several conditions under which more than one hair emerges from a common opening, these conditions and their differential characteristics have been listed in Table 1.

In instances 1. and 2. the multiplicity of hairs results from the successive formation of single hairs on the same papilla. In physiologic hair change, the old hair often remains in the follicle until it is loosened and pushed out by the new hair. Occasionally it may remain long enough for both hairs to be visible at the same time (Koelliker (1), F. Pinkus (2)). Trichostasis spinulosa probably is just an exaggeration of the normal cyclical activity of the papilla and results from the retention of the dead hairs in the hyperkeratotic follicle. These hairs may be embedded in pockets of the follicular wall, and it was suggested (Hochstetter (3), Mitchell (4)) that multiple papillae are present from which these hairs grow. This mechanism, however, has not been substantiated and is not very likely (Corson (5)).

Instance 3., compound follicles, are a matter of common occurrence on the scalp and elsewhere due to the phylogenetic arrangement of the hairs in groups of threes and fives. Koelliker (6), Schulin (7), and Rabl (11) dealt with this phenomenon of several hairs being contained in the same follicle†, Koelliker taught that accessory follicles grow down from the neck of the central one. Rabl apparently favored the view that the superficial parts of independent follicles merge secondarily. However this may be, the fact remains that only the superficial parts

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† Other observations of Koelliker (1), Wertheim (8), and Goette (9) which have been quoted in this connection by Martinotti (10) probably were instances related to trichostasis spinulosa.

of several follicles form a common infundibulum. The deep parts and the papillae are independent units. This almost normal phenomenon has not found much attention in recent texts, and thus it could happen that Loewenthal (12) described it as a new discovery in 1947. He tried to differentiate between "grouped hairs" which emerge close to each other and false and true "compound hairs" depending on how far down in the skin the follicles merge. This, however, seems to be only a difference in degree. Besides, the term "compound hairs" appears inappropriate, as it is the follicles and not the hairs which form a compound. The hairs remain completely independent. It would seem better to speak of "compound follicles" (Koelliker (6): *zusammengesetzte Follikel*) in this connection.

TABLE 1

Instances in which more than one hair emerges from a common opening

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1. Physiologic Hair Change.
old and new hair may be present simultaneously.
 2. Trichostasis spinulosa (*Thysanothrix*).
several or many short dead hairs are retained in the follicle.
 3. Compound Follicles.
the necks of two or more follicles merge and open in a common infundibulum.
 4. Congenital Sinuses and Cysts.
 - a. Preauricular Sinuses
 - b. Pilonidal Cysts
 - c. Dermoid Cysts
many hair roots may spring from the epithelial wall and the hairs may emerge through an outside opening.
 - d. Folliculomas
several more or less well formed hair roots may open into a central cyst.
 5. Multiple Hairs (*Pili multigemini*).
 - a. True Multigeminate Hairs
a composite papilla forms several hair shafts which are separated from each other by the internal root sheath and are enclosed in a common outer root sheath.
 - b. Composite Hairs
a composite papilla forms several partly merged hair shafts.
 6. *Fragilitas Crinium*.
longitudinal splitting of the free end of the hair may extend into the follicle.
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Instance 4., sinuses and cysts bearing hairs, form an interesting chapter of cutaneous and surgical pathology, but need not be discussed here. In them, the lining stratified epithelium which is equivalent with epidermis may form several to many hair follicles from which spring a corresponding number of individual hairs. There are some rare cystic tumors, usually small, possibly related to trichoeplithelioma, in which the tumor matrix may form several more or less well developed hair roots and papillae. These tumors may be termed folliculomas.

Instance 5. is the subject of the present discussion. Instance 6., longitudinal splitting of the hair shaft from the free end, will be mentioned in differential diagnosis later on.

MULTIPLE HAIRS. HISTORY AND CASE REPORTS

Except for a short reference in Savill's book (13) no definite mention of multiple hairs was found in recent literature or in textbooks, but I do not claim that my search was complete. The first one to describe a multiple hair was Flemming (1883) (14) who gave a lucid account of a triplet hair found by chance in sections of bearded skin which had been prepared for histologic teaching. Giovannini (1892) (15) reported a twin hair from the scalp of a child, also an accidental finding in sections prepared for other purposes. Giovannini (16) later published a series of papers on various abnormalities of hairs of the beard, but was more interested in the structure of the papilla than in the condition of the hair itself. He found as many as five shafts surrounded by a common root sheath.

My first case was in a white man 28 years of age who sought treatment for folliculitis of the beard. The infection was confined to a fairly well circumscribed area of the chin. Among the hairs of the chin, there were several which looked three to four times thicker than the average. A magnifying glass revealed that either the ends of the hairs were frayed, or that several closely bundled hairs were present in one follicle. It required considerable force to pull out one of these hairs. They came out as a whole, surrounded by a common sheath. Direct microscopic examination of one of these hairs showed that it consisted of six shafts which converged toward the base, but did not fuse. They appeared to arise from a common very broad bulb which was subdivided and surrounded with a common sheath. The aspect was similar to the several daughter bulbs of tulips or daffodils which sit on a common base and are held together by the sheath of the old bulb which they replaced.

Two hairs were removed by punch biopsy, and transverse sections were made. Fig. 1 shows a cross section of one of the follicles. Enclosed in the outer root sheath, there are seven hairs, septuplets, each surrounded by its cuticle, each showing cortex, and most of them medulla, but in variable shape and distribution. The shafts are surrounded and separated from each other by layers of the internal root sheath. This is even better visible in Fig. 2 which represents a section stained with acid orcein and Giemsa solutions. Huxley's layer is stained deep blue and is seen to be present around and in between the individual hairs. Henle's layer forms a common ring around the whole bundle. Fig. 2 also shows a neighboring hair which has normal dimensions, but has a horse shoe shaped cross-section with Huxley's layer extending deep into the fissure.

The septuplet hairs appear analogous to uniovular twins where each has its own amnion, but both have a common chorion and placenta.

No other cases were observed in more than three years' time until a few months ago when four unusually thick hairs were seen in the beard of a white man 34 years of age who had alopecia areata of the beard. Three of the hairs in question were close to a bald area on the chin, the fourth one was on the neck. Epilation required considerable force and was painful. Microscopic examination of one of the hairs showed it to be similar to those of the first case. Fig. 3 A is the reproduction of a camera lucida drawing because it proved impossible to obtain satisfactory photographs. Several shafts are present which diverge distally and con-

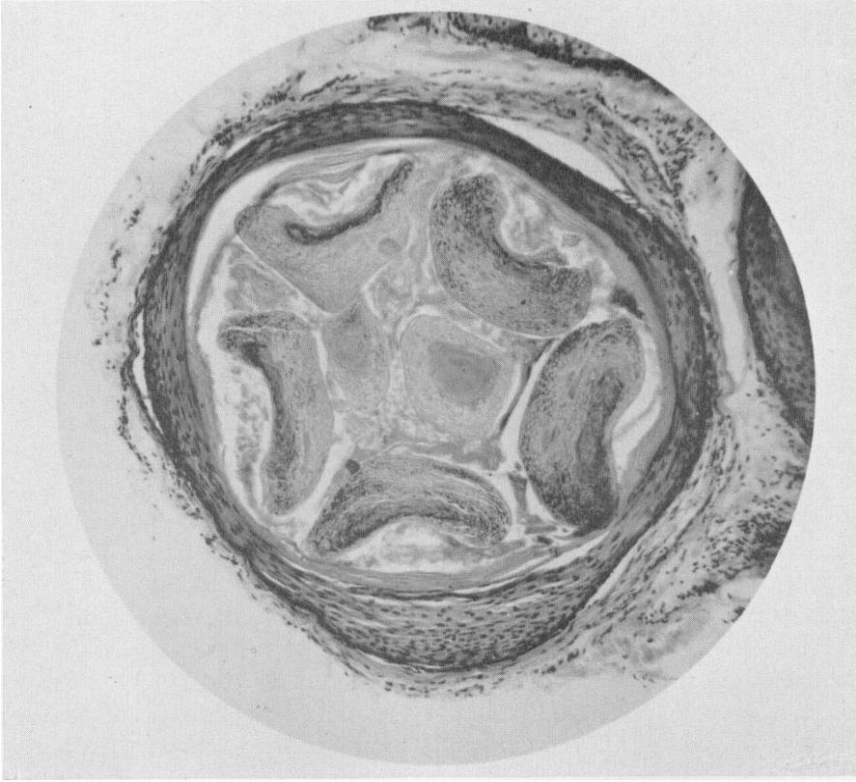


FIG. 1. Crosssection through the middle of the follicle of a multiple hair from Case 1. 90 X Hematoxylin and Eosin. Seven separate hair shafts of uneven shape and size are enclosed in a common outer root sheath and are separated from each other by layers of the inner root sheath.

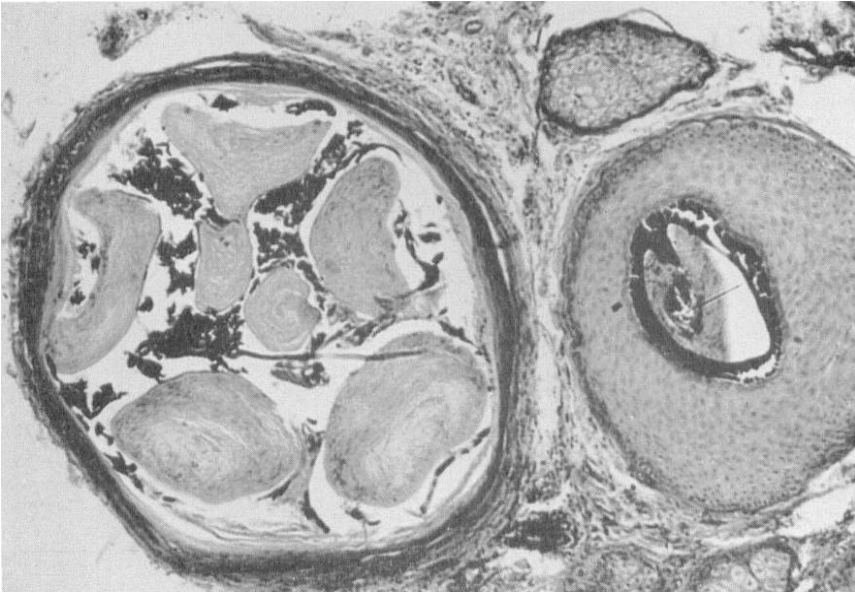


FIG. 2. Crosssection of the same hair as in Fig. 1 and of a neighboring hair of normal diameter. Acid orcein and Giemsa solution. The dark material between the hair shafts is Huxley's layer which stains deep blue with Giemsa solution. The crosssection of the other hair has horse shoe shape, and Huxley's layer extends deep into the fissure.

verge toward a subdivided, composite papilla, as described by Flemming and Giovannini. The second hair (Fig. 3 B) is somewhat different. Division is much less complete. One has the impression that the papilla functioned in different fashion at different times and formed branching and reuniting partial shafts. This may be compared to the twin monsters of embryology.

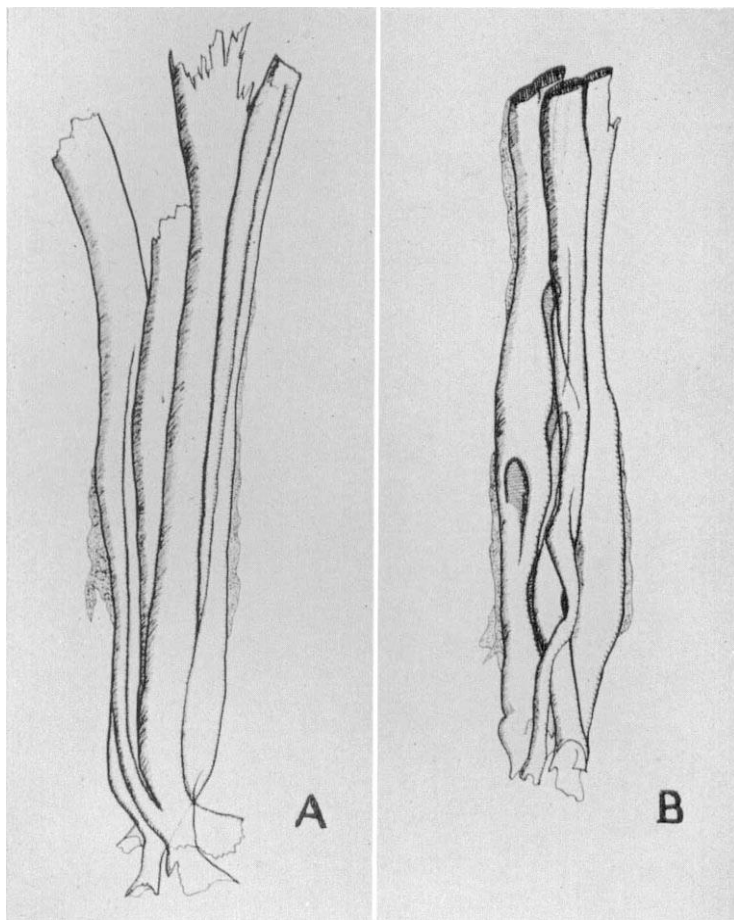


FIG. 3. Semidiagrammatic drawing of epilated hairs of Case 2. A. Almost completely separated hair shafts converge toward a composite papilla. B. Composite papilla has formed alternately fusing and dividing shafts. Note that the number and site of the subdivisions changes from level to level.

OCCURRENCE AND PATHOGENESIS

As with other abnormalities of hair, there is a question if the occurrence of multiple hairs actually is so rare, or if it is just being overlooked. I believe that this anomaly really is uncommon. At a time when anatomists and dermatologists were intensely interested in hairs, there was a lapse of nine years between Flemming's and Giovannini's first observations. Giovannini found only a few cases within the next twenty years, and apparently no other observations have been published. F. Pinkus told me that he had never seen a case of multiple hairs, and I myself have not been able to find additional ones.

Minor aberrations from the norm which furnish the basis for actual subdivision of the hair shaft are not so rare. Flemming (14) already pointed out that several other hairs in his sections had composite papillae and irregular shape of the cross-section of the hair shaft. He assumed an individual disposition to malformation of the papilla. The site of predilection is the bearded chin. Giovannini found various degrees of subdivision of the papilla rather frequently in this region. Von Brunn¹⁷ and Danforth¹⁸ reproduced a number of angular and fluted crosssections of beard hairs in their publications. Koelliker⁶, Giovannini¹⁶, and Danforth¹⁸ mention multiple medullae in individual hairs of the beard. With the exception of Giovannini's first case, all multiple hairs have been reported from the beard, usually from the chin.

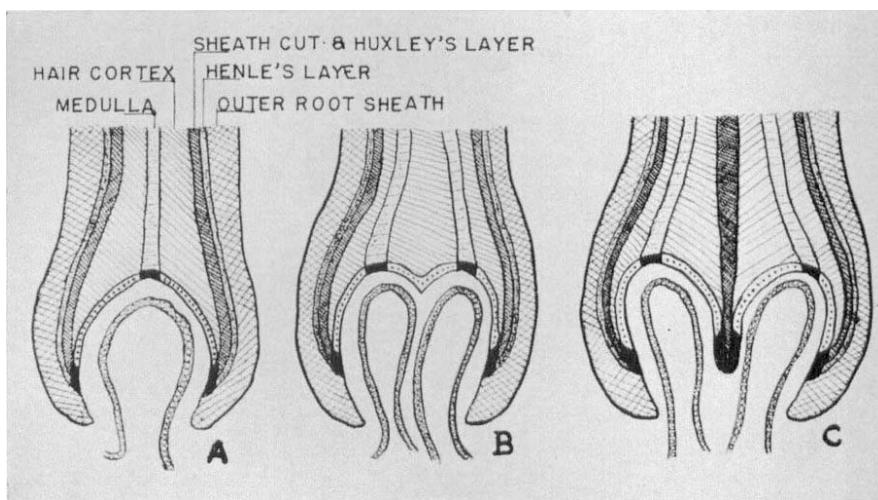


FIG. 4. Diagram of the morphogenesis of a multiple hair. A. Normal hair. The principal layers are indicated. The basal cells of the medulla and of the internal root sheath which produce trichohyalin and "soft keratin" are shown black, the basal cells of the hair cortex which produce "hard keratin" without the formation of trichohyalin are indicated by dots. One capillary loop is shown in the papilla. B. Bimedullated hair resulting from doubling of the tip of the papilla. C. Twin hairs resulting from deep split in the papilla which extends down to the neck. Inner root sheath interposed between the two shafts.

Flemming clearly stated the conditions necessary for a subdivided papilla to produce a divided hair. The different layers of the hair and inner root sheath are formed by concentric zones around the circumference of the papilla (Fig. 4 A). Thus, if only the tip of the papilla is doubled the hair will have two medullae (Fig. 4 B). A completely subdivided hair shaft results only if the split extends down to the neck of the papilla where normally the layers of the inner root sheath are formed (Fig. 4 C). Flemming also discussed the possibility that multiple hairs may be due to partial merging of several papillae rather than to division of one papilla. He favored the second mechanism, and this is indeed the most likely one as all stages of splitting of the papilla were observed by Giovannini.

In another point Flemming apparently erred. He observed partial fusion of the three shafts of his case into a trefoil shape in the upper parts of the follicle, and he explained that this was to be expected when the hairs touched each other

after the inner root sheath had disintegrated as it normally does above the opening of the sebaceous gland. A secondary union of the completely cornified hair is, however, unthinkable. Giovannini observed several bridges between the two shafts in his case, several quite long and composed of cortex and cuticle of the hair. My second case showed alternate fusion and separation of the shafts, and, what is important, the splitting did not always occur in the same manner. There can be only one explanation for these findings. The hair matrix apparently is in a labile state and functions differently at different times. Sometimes one common or a few large shafts are formed, sometimes several thinner ones, and as the hair is pushed out and hardens, it reflects in its cross-section the functional state of the matrix at the time of its formation. There is then no absolute distinction between the completely divided multiple hairs and the partly merged composite hairs. We may consider the latter as a special case of the former, as in embryology double monsters are considered to be incompletely separated twins.

The concept of a labile state of the hair matrix raises an interesting and really fundamental question. A look at the diagram in Fig. 4 shows that if the matrix functions at one time as in C, another time as in B, and later one again as in C, two mechanisms could be responsible. Either the cells in the angle between the two hairs form root sheath cells at one time, hair cortex cells at another time. Or the papillae become temporarily more fused and crowd out those cells which form root sheath. Both possibilities offer considerable difficulties. The formation of the "hard keratin" of the hair cortex which takes place without the intermediary step of trichohyalin formation, and the formation of the "soft keratin" of the highly specialized cells of sheath cuticula and Huxley's layer are so different morphologically and biochemically (cf. Giroud and Leblond (19)) that it is difficult to assume that the same cell could function either way at different times. On the other hand, if the matrix material for the root sheath is crowded out by closer fusion of the papillae, how is it regenerated later when the papillae separate again enough to form several shafts? Sheath matrix cells might migrate around from the peripheral parts of the papilla. In either case it would appear that the actual topographical location of a basal cell, its relative distance from the tip and the neck of the papilla, is the most important determining factor for its specific function. This touches on fundamental problems of determination in embryologic development.

Differential Diagnosis

Table 1 gives a survey of conditions which might enter in differential diagnosis. Most of these were discussed earlier in this paper. The only one that needs additional mention is instance 6, fragilitas crinium with longitudinal splitting of the hair shaft.* Usually the split is restricted to the distal ends of long hairs, but Duhring (22), Parker (23), and Hyde (24) described three cases between 1878 and 1888 in which the split extended into the follicle close to the bulb. The epilated

* The terminology of this condition varies. Longitudinal splitting of the hair is designated as trichiptylosis in French²⁰ and German²¹ texts while the term trichoptilosis is synonymous with trichorrhhexis nodosa in American usage.

hairs, all of them of the beard, apparently resembled multiple hairs fairly closely. The differential points are these: The hairs were not scattered in the beard, but occurred in patches which increased in size over the years. They gave rise to inflammation and considerable discomfort. They could be pulled without pain, and their root is described as atrophic. No mention is made of multiple papillae which are so conspicuous on our hairs. My first patient had folliculitis barbae, but this did not seem to affect the multiple hairs more than the normal ones. All three authors state that the disease caused symptoms only when the men let their beards grow, and that close shaving appeared to be the most helpful therapy. This may account for the fact that apparently no similar cases have been observed in our clean-shaven age.

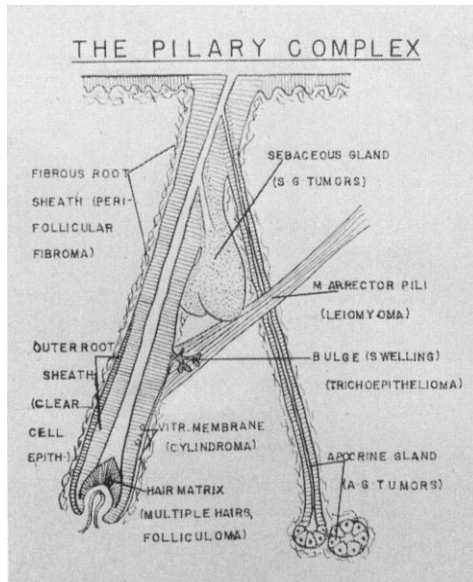


FIG. 5. Diagram of the hair follicle and its accessory and supporting structures, the Pilary Complex. The names of cutaneous tumors which theoretically may be derived from the various parts of the adult pilary complex are shown in parentheses. For details see text.

Comparative Anatomy

It has been mentioned that multiple medullae in individual hairs of the beard are much more common than multiple hairs. Multiple medullae are the rule in some thick animal bristles, for instance in those of the pig and the elephant, and they are associated with composite papillae (Moebius (25), Toldt (26), Kraenzle (27)). Pigs' bristles are described as having multiple brush like free ends, but Kraenzle, in contrast to some older authors, concluded that this brush like end is analogous to fragilitas crinium and is only indirectly due to the peculiar shape of the papilla which may have as many as 16 to 18 pointed tips. This, according to Kraenzle, causes the architecture of the cortex to be less homogeneous and more prone to split.

COMMENT

Flemming's description of a triplet hair and Giovannini's painstaking investigations of multiple hairs and composite papillae have been practically forgotten. My discussion is presented not so much with the purpose of reviving knowledge of a rare anomaly of hair and thus add another item to the album of dermatologic oddities, but it seems to me that multiple hairs deserve interest for other reasons.

One reason was mentioned above. That is the fundamental question of what makes some of the hair matrix cells form cortex, others medulla, cuticle, or root sheath. My patients unfortunately were not available for more extensive studies. Perhaps investigation of other cases if they can be found would shed light on this question. It may also be possible to find out if the same papilla will again form a multiple hair after epilation of the first one.

Another reason for this presentation is that these cases show abnormal potentialities of the papilla and hair matrix which border on the truly pathological, and pathological in this connection means tumor formation. There is at present a strong tendency to attribute all those cutaneous tumors that in their structure exhibit resemblance to adnexal tissues to the activation of embryonal rests, to the "hair germ" of classical anatomy, now usually called the "primary epithelial germ" (Lever (28)). This hypothesis, based on Cohnheim's (29) views, is attractive, and is valuable in the explanation of certain tumors, but it should not let us forget other possibilities. The possibility that adult structures possessing proliferative activity may give rise to tumors cannot be dismissed lightly. The hair and its related and supporting structures, the pilary complex, which undergoes cyclical growth changes all through life appears to offer particularly good possibilities for abnormal trends which may lead to tumor formation. A study of the minor abnormalities of the adult pilary complex may have interesting results. As a matter of fact if one pays attention to such abnormalities in routine biopsy material, a surprisingly large number is encountered. Some of these details will be presented in a later communication. For the present, Fig. 5 lists a number of theoretical possibilities for derivation of tumors from definite single parts of the pilary complex. Some of the possibilities are obvious. Short mention should be made of two parts of the hair root to which little attention has been paid in recent years and which are barely, if at all, mentioned in the textbooks. One of these is the vitreous membrane of the lower part of the follicle which may form hyaline pearls indistinguishable from those found in cylindroma. The other one is the "bulge" (swelling, German: *Wulst*), a structure more obvious in embryonic development, but present on each adult follicle. This epithelial cushion is the site of insertion of the arrector muscle. It may form branching proliferations which may enclose horn pearls and which resemble very much the structure of trichoepithelioma.

SUMMARY

Two cases are presented of multiple (up to septuplet) hairs growing on composite papillae within a common outer root sheath.

Based on descriptions of a triplet hair by Flemming, and of twin and quin-

tuplet hairs by Giovannini, the name of "pili multigemini," multiple or multigeminate hairs, is proposed for this rare abnormality of hair formation.

Occurrence, pathogenesis, differential diagnosis, and comparative anatomy are discussed.

Some thoughts are offered on the significance of minor aberrations of parts of the adult pilary complex for the problem of tumor formation.

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DISCUSSION

DR. WILSON: As you all can readily see I cannot be classed as a fur bearing animal, and I just wanted to make available to Dr. Pinkus, if he hasn't had them, some of my dead chinchilla hides. It was my information, probably wrong since I have listened to his erudite discussion, that not one, not two, not seven, but as many as sixty hairs emerged from one follicle in that animal. I do have such material available, if he would like to investigate it.

DR. PINKUS: I thank Dr. Wilson for his discussion and I gladly accept his offer as I would like very much to add sections of chinchilla skin to my collection. I mentioned chinchilla on purpose because it is a widespread belief that the many hairs of the hair tufts of this animal spring from a common root. However, Dr. Harry H. Wilcox of the Department of Anatomy, University of Pennsylvania, exhibited sections of chinchilla skin at the 62nd annual meeting of the American Association of Anatomists (*Anat. Rec.* **103**: 172, March 1949) and showed that, although as many as 50 hairs exit from the skin through a single pore, each hair is formed in a simple individual follicle with a simple papilla. Thus the chinchilla fur offers another instance of compound follicles rather than of multigeminate hairs.