

**Case Report** 

# Scrotal Hair in Infancy: Outcome Verification of an Isolated Condition Needing Minimal Assessment

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Received date: May 17, 2018; Accepted date: June 04, 2018; Published date: June 15, 2018

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#### Abstract

Rarely, infant boys present with a few, pigmented scrotal hairs and often laboratory assessment is done. This report aims to ascertain any possible pathology of this condition by providing outcome data for 8 patients. This report reviews the clinical history of eight male infants, who initially presented with scrotal hair growth without other signs of virilization. Follow-up documented no progression or recurrence or evidence of androgen excess among 4 of the 8 patients beyond infancy and 4 patients beyond the age of puberty. Only one did not show regression of pubic hair during infancy, but persistence to age 7 when hormone levels were shown to be normal. Hence, these data, plus a summary of the published literature that confirms that this finding, without other evidence of androgen excess, is benign and self-limited and does not require laboratory assessment, but only clinical follow-up.

Keywords: Scrotal hairs; Androgen; Infants

condition and suggests that, while clinical follow-up is indicated, no hormonal evaluation for these patients is needed.

# Introduction

Scrotal hair in infancy is a rare condition that is concerning for parents and doctors alike. It is evidence of excessive androgen stimulation or responsiveness. If there is an underlying pathologic etiology, there will be persistent and excessive androgen and this early evidence would be accompanied by other evidence including genital growth and an accelerated growth rate. Limited information on the natural history of this entity suggests that further evidence of masculinization does not occur. This report further explores whether this is a benign state that, when isolated, does not progress and usually regresses. The aim is to review the characteristic of this finding among the patients presented, and compare with data from a review the literature. These data, together with published reports of 41 other patients [1-11] support the position that this is a benign transient

Retrospective analyses of medical record charts has been done for eight patients who were seen by one or more of the authors and presented with scrotal hair in the first 6 months of life with follow-up

**Cases and Methods** 

presented with scrotal hair in the first 6 months of life with follow-up information regarding persistence of hair, and other growth and development. Five of these patients had hormonal measurements of adrenal and testicular steroid or gonadotropin levels at the age of presentation and one was evaluated at age 7 because scrotal hair had persisted but not progressed. The Hershey Medical Center Human Rights Committee indicated that this chart review did not require IRB approval since patient data cannot be identified. Eight healthy male infants presented a history of having scrotal hair noted between two and six months of age (Table 1).

S No.	Age onset (mos)	Age assessed (mos)	No. of hairs	Progression	Persistence beyond 1 yr	Hormone assessment/Normal for age	Latest follow-up age (yrs)
1	4	5	2	No	No	Yes/yes	4
2	2	6	<10	No	No	Yes/yes	21
3	<6	12	<10	No	Yes	Yes/yes	11
4	5	6	Few	No	No	Yes/yes	12
5	3	3	<10	No	No	No	13
6	Birth	6	3	No	No	No	4
7	3	9	Several	No	No	Yes/yes	15
8	6	8	15	No	Yes	No	2.3

 Table 1: Natural history of scrotal hairs of infancy among 8 boys.

Median age of noted onset was 3 to 4 months (range birth to 6 months); median age of assessment was 6 months (range 3 to 12 months). All infants had a normal prenatal history, including no evidence of maternal androgen excess and a normal neonatal period with normal growth and development. Development and size of penis, testes, and scrotum were also normal. Numbers of hairs were few, ranging from two to 15. In all cases there was no other evidence of inappropriate virilization, excessive linear growth, or further pubic hair growth. There was no history of males in the family having unusual amounts of sexual hair, or females having hirsutism or PCOS. Racial/ethnic categories included eastern and western Europe and Mediterranean.

Scrotal hairs were pigmented; 1.5 cm or longer in length and most were curly. These hairs were documented to regress completely in six of the infants. In two patients, it persisted without progression throughout the observation period, to 28 months and seven years respectively. This latter patient was assessed at seven years of age and found to have normal androstenedione and 17-hydroxyprogesterone levels and a skeletal age of 6 years. This follow up, 2 others into prepubertal years and 4 to or beyond the age of puberty demonstrated normal growth and development.

Hormone analyses were done in a total of 5 patients with results within the normal range for age. Sampling included adrenal hormones before and after ACTH stimulation testing, testosterone, luteinizing hormone, and follicle stimulating hormone. Based upon these normal results, the 3 subsequent patients did not have testing except for clinical follow-up.

## Discussion

Typically when unusual signs of virilization occur before puberty, there is an assessment for pathologic causes. However, in the instance of isolated scrotal hair with no other genital or testicular changes or linear growth acceleration, as the 8 patients reported here, together with 41 cases reported in the literature [1-11] all suggest this clinical presentation is a benign self-limited condition. With the exception of one boy whose mother indicated hairs were present at birth, age of onset was noted for 37 at a median of 4 months (ranging from birth to 7 months), and documented for all 49 between 3 and 10 months (median 6 months (Table 2). The numbers of hairs, which were few, were reported among 20 ranging from 2 to 20 (median 7). Resolution was documented among 33 between 8 and 17 month (median 11 months). Thus, this apparently benign variation with onset during the first 7 months of life is self-limiting. The one patient with persistence, who had hormonal testing at 7 years of age, can be considered to have a diagnosis of premature adrenarche. This suggests a benign condition so that those presenting with no other concerning or abnormal findings on history or physical exam do not need hormonal testing unless the scrotal hair progresses or other signs of virilization develop. Among those with follow-up up to 21 years, no growth or pubertal developmental abnormalities were noted.

Overall, these outcome data suggest that this is a self-limited condition that usually resolves before 12 months of age. According to our experience with the eight patients plus the 41 previously reported

suggest that watchful waiting without hormonal testing is a prudent justified approach for those presenting with isolated scrotal hair without other signs of virilization or linear growth excess. Based on the position that this finding might be the first evidence of a pathologic cause of adrenal or testicular origin, some previous reports have indicated that hormone levels should always be assessed. Hormone testing is reported among 38 in the previous literature plus the 5 in the current report (43 of the total of 49) (Table 2).

	Units	No. of cases	Range	Median
Onset	mos	38	Birth-7 mos	4
Assessment	mos	49	3-10	6
Resolved	mos	33	8-17	11
Hairs	Number	20	2-20	7
Testosterone	ng/dL			
3.5-6 mos		22	8-92	40
7-10 mos		7	<2-11.5	4
17-OH prog	ng/dL			
3.5-6 mos		25	15-160	62
7-10 mos		8	11-81	58
Androstenedione	ng/dL	20	6-93	29
DHEA Sulfate	µg/dL	23	<15-40	30
LH	mIU/mL	11	0.07-2.00	0.4
FSH	mIU/mL	10	0.6-2.0	1.2

Table 2: Data from 41 published cases [1-11] and this report.

Further evidence that this is a benign condition is based on the fact that none of these results identified clearly abnormal levels, with a possible exception of the DHEAS. DHEAS levels, generally fall during infancy, alone are a marker of adrenal androgen secretion from the adrenal reticularis or the fetal adrenal. Elevated levels concomitant with other intermediate metabolites or androgens from the adrenal per se do not indicate a pathologic condition and may be consistent with slow regression of the fetal adrenal zone. The appearance of scrotal hair at this age may result from neonatal adrenal androgen production related to unusual persistence of the fetal adrenal zone beyond birth [12]. Both this potential etiology as well as the increased testosterone secretion during the interval of increased hypothalamic-pituitarytesticular activity of infancy; sometimes referred to as "mini-puberty" of infancy peaks before the age of hormone sample for the individuals reported [13]. Screening for pathologic causes by measuring a variety of hormone levels was pertinent in 33 individuals (Table 2). These levels are within the normal range with the possible exception of DHEAS. Although various assays were apparently used, including some that were not as sensitive as others, the levels extend into the range that has been interpreted to be evidence of persistence of production by the fetal zone of the adrenal gland. This is consistent with a report [14] of serum steroid levels in infants with genital hair, although not specified as scrotal hair, were not different from those without androgen-stimulated changes except half had dehydroepiandrosterone sulfate levels greater than 15 micrograms/dL, consistent with the persistent fetal adrenal zone. Among the 23 levels reported (Table 2), 8 were less than 15 ug/dL, with the remainder being within the range suggesting persistent fetal adrenal zone secretion (depending upon the specificity of the assay). Also, all values of 17hydroyxprogesterone were within the normal range for age with a possible single exception, which was considered not to be indicative of congenital adrenal hyperplasia [8].

Scrotal hair growth could also be hypothesized to relate to increased responsiveness of the androgen receptors in the hair follicles of scrotal skin, either related to racial/ethnic differences or individual variation during infancy. It is appropriate to point out that after infancy, when a boy is exposed to the levels of testosterone characteristic of the active period of infancy; that androgen responses such as sexual hair occur. Hence, in the usual infant male, there must be a relative resistance to the pubic hair promoting properties of the androgen receptor and among those who develop scrotal hair, the receptor is more sensitive to the androgen levels characteristic of the brief period of increased hypothalamic-pituitary-testicular activity of infancy.

This finding may be comparable to benign premature thelarche of infancy which is assumed to result an enhanced responsiveness to low levels of normal ovarian estrogen production during infancy. In the infant female presenting with isolated breast development, benign premature thelarche usually resolves and infant males presenting with isolated scrotal hair also usually resolves, hence, both can be observed without further investigation unless progression is noted.

## Conclusion

The characteristics of the patients presented in the case report, together with the data from previously reported patients indicate that in the absence of other signs of virilization, the otherwise healthy infant boy with isolated scrotal hair can be conservatively managed without hormonal testing. Progression does not occur and, in most cases, resolution by one year of age can be anticipated.

## **Conflict of Interest**

The authors have no conflicts of interest relevant to this article to disclose.

## **Financial Disclosure**

The authors have no financial relationships relevant to this article to disclose.

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