Coccygeal Polypoid Eccrine Nevus Presenting as a Skin Tag: Case Report and Review of the Literature

Christopher G Gibson[1], Amy M Zidron[2], Ruby S Gibson[3], Francis Essien[4] and Scott A Jenkinson[5]

[1]Office of Graduate Medical Education, Fairfield Medical Center, Lancaster, OH, USA. [2]Department of Pediatrics, Heritage College of Osteopathic Medicine, Ohio University, Athens, OH, USA. [3]Long School of Medicine, University of Texas Health San Antonio, San Antonio, TX, USA. [4]Keesler Air Force Base, Biloxi, MS, USA. [5]Department of Specialty Medicine, Heritage College of Osteopathic Medicine, Ohio University, Athens, OH, USA.

Clinical Medicine Insights: Case Reports
Volume 12: 1–3

DOI: 10.1177/1179547619853534
SAGE

ABSTRACT: Coccygeal polypoid eccrine nevi (CPEN) are rare, benign, cutaneous polypoid lesions localized to the coccyx region that are characterized by areas of hyperplastic eccrine ducts without hyperhidrosis. We present the case of an asymptomatic 16-month-old female with a congenital lesion in the lower sacral area and review the literature and the differential diagnosis for CPEN.

KEYWORDS: coccygeal polypoid eccrine nevus, coccyx, hamartoma

RECEIVED: May 1, 2019. **ACCEPTED:** May 4, 2019.

TYPE: Case Report

FUNDING: The author(s) received no financial support for the research, authorship, and/or publication of this article.

DECLARATION OF CONFLICTING INTERESTS: The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

CORRESPONDING AUTHOR: Christopher G Gibson, Office of Graduate Medical Education, Fairfield Medical Center, 401 N Ewing Street, Lancaster, OH 43130, USA. Email: drcgibson@fmchealth.org

Introduction

Eccrine nevus is a rare, benign, congenital hamartoma composed of hyperplastic eccrine glands commonly found on forearms with associated hyperhidrosis.[1] Although there have been isolated reports of eccrine nevi associated with hypopigmentation, plaques, brown papules, and depressed lesions, eccrine nevi usually do not have underlying skin findings.[2-5] An unusual variant of eccrine nevi consisting of coccygeal polypoid lesions without hyperhidrosis has been termed coccygeal polypoid eccrine nevus (CPEN).[6,12] We present the case of an asymptomatic 16-month-old female with a congenital lesion in the lower sacral area. Few cases have been reported in the medical literature.[7-9] The literature shall be reviewed in conjunction with the differential diagnoses that should be considered when encountering a sacral lesion.

Case Report

A 16-month-old female was brought to the clinic for removal of an asymptomatic papule in the coccygeal region that was present since birth. The patient was otherwise healthy with no significant birth defects or family history of similar lesions. The lesion was not associated with any pain, tenderness, constitutional symptoms, localized hyperhidrosis, or hypertrichosis. Grossly, the lesion was a solitary 0.3-cm, gray-tan, firm, nonerythematous, nonblanching papule on the patient's lower coccyx, and a sacral X-ray demonstrated a normal 5-segment sacrum. A shave biopsy revealed a polypoid lesion with a nonacanthotic epidermis and a normally vascularized fibrous dermis. A focal collection revealed numerous hyperplastic eccrine ducts. A pilar structure is also noted at the pedicle of the lesion most likely a part of the hair

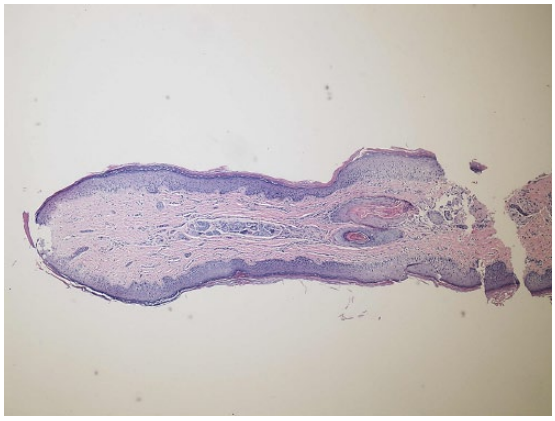

Figure 1. Elongated skin lesion covered with a thin layer of epidermis and normally vascularized dermis. It demonstrates the central coiled eccrine gland and surrounding dermis (hematoxylin/eosin stain 40×).

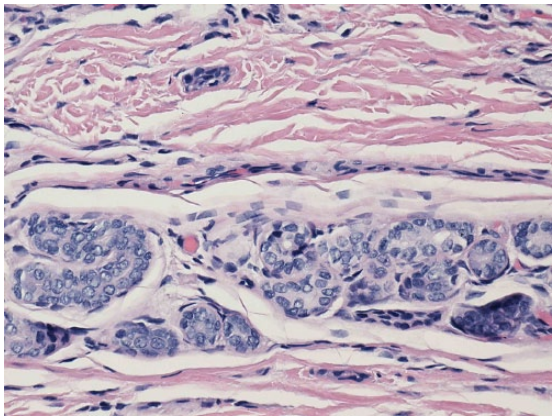

Figure 2. Hyperplastic eccrine glands surrounded by fibrous stroma (hematoxylin/eosin stain 400×).

Table 1. Differential diagnosis of sacral skin lesions.

DIFFERENTIAL DIAGNOSIS	DEMOGRAPHICS	HISTOLOGY	HYPERHIDROSIS	PAIN	DESCRIPTION	LOCATION
Eccrine nevus	F=M Congenital to adolescent	Proliferation and hyperplastic eccrine sweat glands No vascular changes	Yes	No	No overlying skin abnormality (occasional depressed nodule or hyperpigmentation/ hypopigmentation)	Upper extremities: forearm and wrist; occasional back/trunk
Coccygeal polypoid eccrine nevus (CPEN)	F>M Present at birth	Hyperplastic eccrine ducts surrounded by adipose tissue; vascular ectasia seen occasionally	No	No	Flesh-colored papule, acanthotic epidermis	Coccygeal
Eccrine angiomatous hamartoma	M=F congenital/early childhood	Deep dermis—Increased number of capillaries and increased number of eccrine glands	1/3 of cases	Yes	Flesh-colored, bluish or reddish solitary papule; pruritus, hypertrichosis	Lower extremities
Acrochordon ("skin tag")	Adults	No hypertrophic eccrine glands, central adipose tissue, loose collagen bundles, flattened epidermis	No	No	Flesh-colored, pedunculated, filiform	Skin folds
Caudal appendage[10]	Congenital	Presence of nerve fibers, striated muscle fibers, or cartilage	No	No	Skin-covered caudal midline protrusion associated with spinal dysraphism and tethered spinal cord	Coccygeal
Lumbosacral lipoma[11]	Associated with spina bifida	Mature fat tissue	No	variable	Growing mass Neurological deficits	lumbosacral

Abbreviations: F, female; M, male.

bearing skin of the sacral area (Figures 1 and 2). The patient had no recurrence of the lesion a year after the excision.

Conclusions

Coccygeal polypoid eccrine nevus is an asymptomatic congenital polypoid lesion with no reported evidence of hyperhidrosis.[6] Histologically, there are hyperplastic eccrine ducts surrounded by adipose tissue and occasional vascular ectasia. There have been few cases of CPEN in the literature, with only 1 case associated with other congenital anomalies.[9] CPEN occurs predominately in women. The lesions ranged from 3 to 7 mm and are associated with adipose tissue.

The differential for CPEN includes eccrine nevus, eccrine angiomatous hamartoma, acrochordon, caudal appendage, and lumbosacral lipoma (Table 1). Histology, clinical presentation, and localization are important in distinguishing CPEN from other sacral skin lesion (Table 1). CPEN should be considered in the differential diagnosis of a coccygeal skin lesion in infants.

Author Contributions

Acquisition of data: Scott A Jenkinson, Amy M Zidron, Christopher G Gibson

Analysis and interpretation of data: Francis Essien, Christopher G Gibson, Ruby S Gibson

Critical revision: Christopher G Gibson, Amy M Zidron, Ruby S Gibson

Drafting of manuscript: Francis Essien, Christopher G Gibson, Ruby S Gibson

Final approval of the version to be published: Scott A Jenkinson, Amy M Zidron, Christopher G Gibson, Ruby S Gibson, Francis Essien

Study conception and design: Scott A Jenkinson, Christopher G Gibson, Amy M Zidron

Ethical Approval

Any identifying information was removed, and the Office of Human Research Protections for Ohio Health and the Institutional Review Board approved of this paper.

REFERENCES

1. Kawaoka JC, Gray J, Schappell D, Robinson-Bostom L. Eccrine nevus. *Am J Derm*. 2004;51:301–304.
2. Salama H, Shwayder T. Eccrine nevus presenting as a hypopigmented patch. *Pediatr Dermatol*. 2008;25:613–615.
3. Tempark T, Shwayder T. Mucinous eccrine naevus: case report and review of the literature. *Clin Exp Dermatol*. 2012;38:1–6.
4. Imai S, Nitto H. Eccrine nevus with epidermal changes. *Dermatologica*. 1983;166:84–88.
5. Morris ES, Scheel MM, Lundquist KF, Raimer SS. Grouped papules on the arm of an infant. *Arch Dermatol*. 2000;136:549552–549552.
6. Alashari M, Torakawa J. True tail in a newborn. *Pediatr Dermatol*. 1995;12: 263–266.
7. Pierre-Kahn A, Zerah M, Renier D, et al. Congenital lumbosacral lipomas. *Childs Nerv Syst*. 1997;13:298–334.
8. Oh SW, Kang TW, Kim YC, Lew W. Coccygeal polypoid eccrine naevus. *Br J Dermatol*. 2007;157:614.
9. Mahdavy M, Smoller BR. Eccrine nevus presenting as a perianal skin tag: a case report and review of the literature. *Am J Dermatopathol*. 2002;24:361.
10. Park MY, Lee ES. Two cases of coccygeal polypoid eccrine nevi presenting as skin tags. *Ann Dermatol*. 2009;21:440.
11. Frouin E, Riviere B, Maillet O, et al. Coccygeal polypoid eccrine nevus associated with imperforate anus and unilateral multicystic kidney dysplasia. *J Cutan Pathol*. 2016;43:697–701.
12. Charifa A, Ko CJ. Coccygeal polypoid eccrine nevus. *Arch Pathol Laborat Med*. doi:10.5858/arpa.2018-0055-RS.