

An Unusual Meningothelial Element in a Hairy Polyp of the Hard Palate

Si-Hyong Jang · Kyueng-Whan Min
Woong Na · Se Min Jang
Seung Sam Paik

Department of Pathology, College of
Medicine, Hanyang University, Seoul,
Korea

Received : September 6, 2007
Accepted : July 25, 2008

Corresponding Author

Seung Sam Paik, M.D.
Department of Pathology, College of Medicine,
Hanyang University, 17 Haengdang-dong,
Seongdong-gu, Seoul 133-792, Korea
Tel: 02-2290-8252
Fax: 02-2296-7502
E-mail: sspaik@hanyang.ac.kr

Hairy polyps are a rare malformations of bigerminal origin that comprise of both ectodermal and mesodermal elements. Meningothelial elements are an extremely rare pathologic finding in hairy polyps. Here we report a case of a hairy polyp with a meningothelial element, which originated from the hard palate. A 1-year-old boy was evaluated for an intraoral mass accompanied by multiple congenital anomalies. A small polypoid mass was noted at the midline of the hard palate. The lesion had central fibroconnective tissue with an unusual stromal component showing reticulated anastomosing pseudovascular patterns. Immunohistochemical staining of the cells lining the pseudovascular spaces and the interstitial cells revealed vimentin and epithelial membrane antigen positivity.

Key Words : Hard palate; Polyp; Meningothelial element

Hairy polyps are rare congenital malformation lesions composed of both ectodermal and mesodermal elements.^{1,2} The lesion was first described in the English literature by Brown-Kelly in 1918,³ and to date more than 150 cases have been documented. Meningothelial elements are an extremely rare histologic finding in hairy polyps. To the best of our knowledge, there has been only one case cited in the English literature.² Here we report an unusual case of a hairy polyp with a meningothelial element.

CASE REPORT

A 1-year-old boy visited our hospital with a chief complaint of a congenital oropharyngeal mass. He was born by spontaneous vaginal delivery at full term. His birth weight was 3,021 grams. On physical examination, the oral cavity exhibited a 0.7 cm sized bean-like pedunculated mass with soft consistency, which originated from the hard palate. Hypospadias of the penis was also noted. Simple x-ray and computed tomography demonstrated a defect of the alveolar bone and an incomplete cleft palate. Magnetic resonance imaging showed no obvious

connection between the palatal mass and the central nervous system. Surgical excision of the mass was performed, and a local flap was made. On gross examination, the specimen was an oval yellowish white solid mass with a smooth outer surface. The cut surface had a homogeneous tan-yellowish white fibrotic appearance. Microscopically, the lesion was covered by the keratinizing stratified squamous epithelium. The submucosal stroma was characterized by sebaceous glands, striated muscle bundles and fibroadipose tissue. The central core was composed of fibroconnective tissue intermixed with unusual stromal tissue with reticulated and anastomosing pseudovascular patterns. The anastomosing pseudovascular spaces were lined by flattened to cuboidal cells and clear polygonal cells. The reticulated areas showed bundles of bland-looking spindle cells, which had elongated or oval to round nuclei and fibrillary cytoplasm. Immunohistochemical staining of the cells lining the pseudovascular spaces and the interstitial cells revealed vimentin and epithelial membrane antigen positivity (Fig. 1). These cells were negative for S-100 protein, cytokeratin, factor VIII-related antigen, alpha-fetoprotein, Ulex europaeus lectin, and glial fibrillary acidic protein.

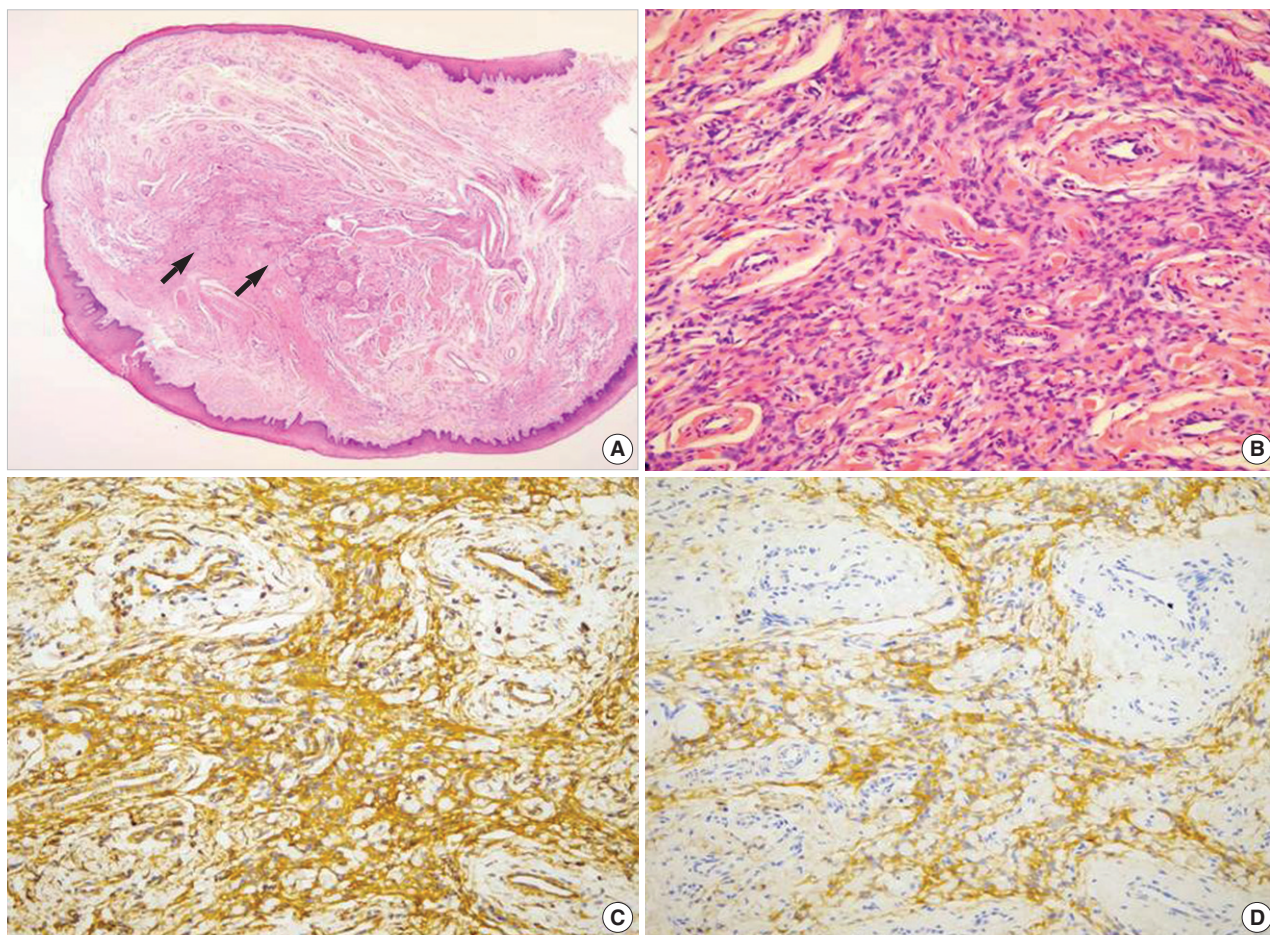


Fig. 1. The resected mass shows polypoid feature with unusual stromal meningothelial element (arrows) (A, B). The cells lining the pseudovascular spaces and the interstitial cells are positive for vimentin (C) and epithelial membrane antigen (D).

DISCUSSION

Meningothelial tissue is a rare histologic finding in a hairy polyp. Some proposed theories of extracranial meningothelial proliferation can be applied to explain the meningothelial element in a hairy polyp. Arachnoid cells in the sheaths of peripheral and cranial nerves can be a source of extracranial meningothelial proliferation.^{4,5} The inclusion theory suggests that germinal layers become displaced in deeper tissue layers, inhibiting normal fusion during embryogenesis and causing development of a mass.⁶ Heterotopic glial tissue in the nasal fossa in the form of a nasal glioma may be another example of displaced neuroectodermal cells.⁷ Totipotent cells escape the normal mechanisms of regulation and control in the embryo and lead to formation of a mass.^{4,8}

The main histologic differential diagnosis of meningothelial elements in a hairy polyp includes endodermal sinus tumors, meningothelial heterotopia, rudimentary meningoceles, and

angiomas. In endodermal sinus or yolk sac tumors, cells with atypical cytologic features, eosinophilic hyaline globules and Schiller-Duval bodies are characteristic.⁹ Negativity for cytokeratin, alpha-fetoprotein, factor VIII-related antigen, and Ulex europaeus can help to exclude endodermal sinus tumors and vasoformative lesions from the differential diagnosis. Unlike hairy polyps, meningothelial heterotopias of the skin and rudimentary meningoceles occur primarily in the subcutis of the scalp.^{10,11}

Surgical excision is the treatment of choice for hairy polyps. However, preoperative evaluation for the presence of intracranial connections is important because encephaloceles and other lesions with intracranial connections may clinically resemble this lesion.¹

Although this histologic entity is extremely rare, recognition and awareness are necessary so pathologists will avoid confusion with other differential lesions.

REFERENCES

1. Franco V, Florena AM, Lombardo F, Restivo S. Bilateral hairy polyp of the oropharynx. *J Laryngol Otol* 1996; 110: 288-90.
2. Olivares-Pakzad BA, Tazelaar HD, Dehner LP, Kasperbauer JL, Bite U. Oropharyngeal hairy polyp with meningothelial elements. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1995; 79: 462-8.
3. Brown-Kelly A. Hairy or dermoid polyp of the pharynx and nasopharynx. *J Laryngol Rhinol* 1918; 33: 65-75.
4. Thompson LD, Gyure KA. Extracranial sinonasal tract meningiomas: a clinicopathologic study of 30 cases with a review of the literature. *Am J Surg Pathol* 2000; 24: 640-50.
5. Sexton M. Hairy polyp of the oropharynx: a case report with speculation on nosology. *Am J Dermatopathol* 1990; 12: 294-98.
6. McShane D, el Sherif I, Doyle-Kelly W, Fennell G, Walsh M. Dermoids (hairy polyps) of the oro-nasopharynx. *J Laryngol Otol* 1989; 103: 612-5.
7. Yeoh GP, Bale PM, de Silva M. Nasal cerebral heterotopia: the so-called nasal glioma or sequestered encephalocele and its variants. *Pediatr Pathol* 1989; 9: 531-49.
8. Chaudry AP, Lore JM Jr, Fisher JE, Gambrino AG. So-called hairy polyps or teratoid tumors of the nasopharynx. *Arch Otolaryngol Head Neck Surg* 1978; 104: 517-25.
9. Dehner LP, Mills A, Talerman A, Billman GF, Krous HF, Platz CE. Germ cell neoplasm of head and neck soft tissues: a pathologic spectrum of teratomatous and endodermal sinus tumors. *Hum Pathol* 1990; 21: 309-18.
10. Suster S, Rosai J. Hamartoma of the scalp with ectopic meningothelial elements: a distinctive benign soft tissue lesion that may simulate angiosarcoma. *Am J Surg Pathol* 1990; 14: 1-11.
11. Marrogi AJ, Swanson PE, Kyriakos M, Wick MR. Rudimentary meningocele of the skin: clinicopathologic features and differential diagnosis. *J Cutan Pathol* 1991; 18: 178-88.