The Role of Fine-Needle Aspiration Biopsy in the Diagnosis and Management of Juvenile Hemangioma of the Parotid Gland and Cheek

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- **Background.**—The current recommendation for the management of juvenile hemangiomas (JH) is to delay treatment in the hope of spontaneous regression. However, accurate diagnosis is necessary before considering conservative management. Traditionally, the diagnosis of JH has required excisional biopsy. The cytology literature on this relatively rare neoplasm is sparse.

**Objective.**—To present our experience with fine-needle aspiration in the diagnosis and management of JH.

**Design.**—Three cases with a cytologic diagnosis consistent with JH of the parotid gland and cheek were identified from our cytopathology files. Aspirate smears, immunohistochemical studies, computed tomographic scan findings, and clinical follow-up were reviewed.

**Results.**—Patients were female infants ranging in age from 3 to 9 months and presented with an oval firm mass (size range, 2.0–5.0 cm) involving the parotid gland (2 cases) and cheek (1 case). Computed tomographic scan with contrast demonstrated homogeneous enhancement. Aspirate smears revealed spindle-shaped cells in sheets and clusters in a background of blood. The parotid gland aspirates and cell block preparations revealed ductal structures entrapped in sheets of spindle-shaped cells. Immunohistochemical studies revealed prominent vascular spaces lined by CD34 and factor VIII–positive flattened endothelial cells. The diagnosis of JH was rendered on the basis of the cytologic findings in conjunction with the radiologic and clinical findings. On clinical follow-up (8–24 months), none of the patients has shown any progression of the lesion.

**Conclusions.**—Fine-needle aspiration, in conjunction with imaging studies, is a useful tool in the diagnosis and management of JH. It eliminates the need for surgical excision for diagnostic purposes and allows for clinical follow-up of patients with JH.

_J_ uvenile hemangioma (JH) occurs principally in patients younger than 1 year.\(^1,2\) Clinically, it may be located on any body surface but most commonly occurs in the head and neck region, particularly the parotid gland. The lesion usually develops by 6 months of age and 70% to 95% of the cases show spontaneous regression by the age of 7 years.

Clinically, superficial JHs usually do not pose much diagnostic difficulty, since they impart a bluish hue to the overlying skin and are associated with thrills and bruits.\(^3,4\) In contrast, deeply seated JH (eg, those involving the parotid gland or cheek) may have no consistent specific clinical signs, which may result in preoperative misdiagnosis.\(^5\) Careful limited resection of the lesions and the involved gland, as advocated by some authors,\(^6\) may have resulted, in part, from the need for distinguishing JH from malignant solid neoplasms of infancy that may be associated with a grave prognosis.

The cytology literature on this relatively rare neoplasm is sparse.\(^7,8\) The purpose of this study is to present our experience with fine-needle aspiration (FNA) in the diagnosis and management of JH. To our knowledge, this is the first report on a series of cases where reliance on the FNA diagnosis of JH resulted in a more conservative approach in the follow-up of these patients.

**MATERIALS AND METHODS**

A search of the cytology files of SUNY Upstate Medical University Hospital, Syracuse, NY, revealed 3 cases with an FNA diagnosis “consistent with JH.” The clinical data, radiologic and cytologic findings, and subsequent clinical follow-up for all these cases were reviewed.

All FNAs were performed by cytopathologists using 25- or 23-gauge (0.25- or 0.23-mm) needles. No analgesia was used. Physical restraint to allow fixation of the head and neck area (site of FNA) was used. In all cases the material obtained was smeared onto uncoated glass slides either air-dried or fixed in 95% ethanol for a Diff-Quik or a Papanicolaou stain, respectively. A cell block was procured by FNA; it was fixed in 10% neutral-buffered formalin, embedded in paraffin, sectioned at 4 \(\mu\)m, and stained with hematoxylin-eosin.

Immunohistochemical studies were performed in selected cases on paraffin-embedded sections by the labeled streptavidin-biotin peroxidase method.\(^9\) The antibodies used included those against vimentin (mouse monoclonal, 1:10; Dako Corporation, Carpinteria, Calif), CD34 (mouse monoclonal, 1:25; Labvision, Freemont, Calif), and factor VIII-AG (1:400, Dako).

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**Clinical Findings, Radiologic Findings, and Cytologic Diagnoses**

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<th>Case No./Sex/Age, mo</th>
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<td>1/F/9</td>
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<td>H/O of parotid mass developing several weeks after birth; initially enlarged and stabilized at 3.0 cm; R/O malignant neoplasm vs hemangioma</td>
<td>A lobular isointense mass in the region of the right parotid gland; appears to be cystic; DDX of hemangioma vs cystic neoplasm</td>
<td>Spindle cell proliferation consistent with juvenile hemangioma</td>
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<tr>
<td>2/F/4</td>
<td>Right parotid gland</td>
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<td>Spindle cell proliferation consistent with juvenile hemangioma</td>
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<tr>
<td>3/F/3</td>
<td>Right cheek mass</td>
<td>Right cheek mass present at birth; has increased in size lately; overlying skin unreMARKable; R/O solid tumor vs hemangioma</td>
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<td>Spindle cell proliferation consistent with juvenile hemangioma</td>
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* CT indicates computed tomography; H/O, history of; R/O, rule out; and DDX, differential diagnosis.

**RESULTS**

The Table summarizes the clinical and radiologic findings and the cytologic diagnoses. The cytologic diagnosis of spindle cell proliferation consistent with JH was rendered in all cases.

In all cases, the aspirate smears were composed of cohesive groups of elongated bland spindle cells with oval bland nuclei in a bloody background (Figure 1, A). Hypercellular groups of spindle cells arranged in compact 3-dimensional coils were also seen (Figure 1, B). The spindle cells exhibited a scant to moderate amount of homogenous cytoplasm. Nuclei demonstrated even distribution of slightly coarse chromatin. No anaplasia, mitosis, or prominent nucleoli were noted. Rare isolated acini and ductal structures were also identified in cases 1 and 2.

Cell block preparations revealed diminutive fragments of tissue comprising solid-appearing areas of spindle cells similar in appearance to cells identified on the aspirate smears. Small open spaces (reminiscent of capillary spaces), lined by uniform plump to flattened cells, presumably endothelial cells, were also identified in some areas (Figure 2, A and B). Ductal structures consistent with salivary gland ducts, admixed with solid areas of spindle cells, were seen in parotid gland aspirates (Figure 2, A).

Immunoperoxidase staining demonstrated positive factor VIII and CD34 staining of uniform plump to flattened cells lining the small oval spaces without staining of the solid spindle cell areas (Figure 3, A and B). The immunohistochemical staining pattern supported the endothelial origin of the uniform plump to flattened spindle cells. Diffuse and strong vimentin expression was noted in all the spindle cells.

In conjunction with the clinical and radiologic findings, a cytologic diagnosis of spindle cell proliferation, consistent with JH, was rendered in all 3 cases. Based on the FNA findings, a more conservative approach was pursued. None of these patients underwent surgical excision. Clinical follow-up of these patients for 8 to 24 months did not reveal any progression of the lesions. Two patients (cases 1 and 2) demonstrated a decrease in size of the lesion at 8 and 12 months of follow-up.

**COMMENT**

Juvenile hemangioma, a distinctive neoplasm with predilection for girls, commonly involves the head and neck region, particularly the parotid gland, in children younger than 1 year.

In contrast to the decided left-sided laterality reported in the literature, all of our cases of JH involved the right side. The treatment of JH in children is conservative, unless surgical intervention is imposed by clinical considerations, since many patients experience spontaneous resolution.

However, it is imperative that a definitive diagnosis is established for effective management of JH. Clinically, JH may enlarge rapidly and simulate an aggressive malignant neoplasm. Based on clinical examination, distinction of deeply seated JH of the parotid gland and cheek from solid tumors of infancy may not be possible. Juvenile hemangioma is also difficult to differentiate from hypervascular tumors based on contrast-enhanced computed tomography and magnetic resonance imaging findings. Hence, in most instances, an excisional biopsy is performed to establish a definitive diagnosis and rule out a malignant neoplasm.

Fine-needle aspiration is being used increasingly in the evaluation of pediatric benign and malignant neoplasms. To our knowledge, only 2 prior cases of FNA of this lesion have been reported in the English-language literature. Hillborne et al described the cytologic findings in a case of surgically confirmed JH with a preoperative FNA diagnosis of low-grade neoplasm. Erhardt et al recently reported on the FNA cytologic findings in a case diagnosed as infantile cellular hemangioma; however, no clinical or surgical follow-up was provided in this patient. In the present series, we report 3 additional cases of infantile cellular hemangioma (JH) that were diagnosed by FNA and managed conservatively.

Our findings of hypercellular and cohesive groups of bland spindle cells arranged in 3-dimensional coils and a bloody background are similar to those of the previously reported cases. However, these findings are nonspecific and cannot be considered diagnostic of JH. In infants, the lesions of head and neck that may be composed of spindle
cells include pleomorphic adenoma, fibromatosis, and uncommon sarcomas.14–19 The predominance of a chondromyxoid background and epithelial cells in pleomorphic adenoma, as well as the pleomorphic spindle cell pattern in sarcomas, allows for distinction of these entities from the bland spindle cells of JH.14,18,19 Differentiation of embryonal rhabdomyosarcoma, with predominantly spindle cells, from JH may pose a diagnostic challenge. In contrast to hypercellular groups of cohesive bland spindle cells, which are seen in JH, aspirate smears of embryonal rhabdomyosarcoma demonstrate highly discohesive hypercellular groups of small round cells and spindle cells that have hyperchromatic nuclei with fine chromatin and inconspicuous nucleoli.16,17

Scant to moderately cellular smears composed of a monomorphic population of loose to cohesive groups of bland spindle cells have also been described in fibromatosis.13 In our experience, aspirate smear findings alone cannot distinguish the bland spindle cells of JH from those of fibromatosis. In our series, cell block preparation and immunostains proved to be most useful in identifying the specific nature of the spindle cells. Cell block sections revealed open spaces within a conglomerate of spindle cells. Factor VIII and CD34 highlighted the endothelial cells lining these spaces, thereby allowing us to categorize the lesion as vascular rather than fibrous proliferation.

The differential diagnosis based on the radiologic findings included hemangioma in all 3 cases. However, the possibility of a hypervascular malignant solid tumor of infancy (2 cases) or an infectious process (1 case) could not be excluded. The cytologic and immunohistochemical findings in conjunction with the clinical findings of a facial mass occurring in infants and the radiologic findings of a possible vascular lesion were considered to be consistent with JH. The patients have been followed up for a period of 8 to 24 months without any progression of their lesion. A decrease in size suggestive of involutional changes was noted in 2 cases.

The possible risk of clinically significant hemorrhage following aspiration of vascular lesions has raised safety concerns about the FNA procedure.20 On the contrary, other reports suggest that the hazards of FNA biopsy of vascular lesions have been overstated.21,22 We did not encounter post-FNA bleeding or hematoma in any of our 3 cases. Bleeding complications were not reported in either of the 2 previously described cases of aspiration cytologic analysis of JH. Nevertheless, we recommend careful observation of infants for evidence of bleeding or hematoma following the procedure, as has also been suggested by Erdhardt et al.8

In summary, FNA cytologic testing, in conjunction with radiologic and clinical findings, is useful in the diagnosis and distinction of JH from other solid tumors of infancy with a predominant spindle cell pattern. Fine-needle aspiration is valuable not only in confirming the benign nature of a mass but also in allowing for a longer period of
observation that may coincide with, or be followed by, its spontaneous regression.

References