CASE REPORT

Parotid gland hemangioma with cytomegalovirus infection
K. R. Chatura1, Mohammad Shahid Iqbal2

1Department of Pathology, Jagadguru Jayadeva Murugarajendra Medical College, Davangere, Karnataka, India, 2Department of Pathology, Kamineni Institute of Medical Sciences, Narketpally, Nalgonda, Andhra Pradesh, India

Abstract
Salivary glands are a common site for hemangiomas in the head and neck region. Surgical pathologists are unfamiliar with hemangioma in salivary glands. We report a rare case of arteriovenous hemangioma of the parotid gland associated with cytomegalovirus.

Keywords
Cytomegalovirus, hemangioma, parotid

Introduction
Hemangiomas are the most common benign vasoformative tumors of infancy and childhood. According to Enzinger and Weiss, hemangiomas are broadly classified into capillary, cavernous and miscellaneous forms such as verrucous, venous, arteriovenous hemangiomas and so forth.[1] Cytomegalovirus (CMV) infection has been observed in the parotid gland of infants.[2] To the best of our knowledge, however, only two cases of parotid juvenile hemangioma associated with CMV infection have been described.[3,4] Therefore, we present a rare case of arteriovenous hemangioma with CMV infection in the salivary gland, hitherto not reported.

Case Report
A 5-month-old male baby presented with a swelling on the right side of his face for the past 2 months. On examination, a mass was noted extending from the right preauricular region to the middle of the neck. Sonography showed a large lobulated tumor in the right parotid region. The left parotid gland was normal. The parotid tumor was resected for a suspected clinical diagnosis of cystic hygroma and sent for histopathological examination. No other clinical abnormalities had been detected. Mother and child were not immunocompromised.

Pathological findings
Macroscopically, a lobulated, soft tumor measuring 4 cm × 3 cm was received. It was unencapsulated. There were areas of hemorrhage but no necrosis. Histological examination did not demonstrate a border between the tumor and parotid gland. Large thick and thin walled vessels filled with blood were seen to intermingle with salivary lobules [Figure 1]. Large eosinophilic intranuclear inclusions with halos (owl’s eye inclusions) and also basophilic cytoplasmic inclusions were seen in many ductal epithelial cells [Figure 2]. Scant scattered chronic inflammatory cells were seen in some lobules. Based on these light microscopic findings, lesion was diagnosed as arteriovenous hemangioma associated with parotid CMV infection.

Discussion
Hemangioma is considered to be the most common neoplasm of childhood, and is one of the most common salivary gland tumors of childhood and infancy, but is rarely biopsied and is, therefore, often unfamiliar to the surgical pathologist.[5] Hemangiomas account for approximately 0.4% salivary gland tumors and occur almost exclusively in the parotid (90%) representing approximately 50% of parotid tumors in the 1st year of life.[6]
Salivary gland hemangioma is relatively rare in surgical pathology files of Armed Forces Institute of Pathology, includes usual hemangioma subtypes, mainly in females, and juvenile hemangioma, displaying distinctive histology predominately in males. Coincidental viral cytopathic effect of CMV infection was identified in only one but not in the case of arteriovenous hemangioma.[5]

Long before the advent of diagnostic virology, pathologists have been aware of the presence of large inclusion-bearing cells in the ducts of salivary glands and in other organs. Contemporary studies have revealed a surprisingly high incidence of these inclusion-bearing or cytomegalic cells in the duct epithelium of salivary glands in infants at necropsy, 12% of 183 necropsies.[7]

In our case, ductal epithelium showed viral cytopathic effect of CMV infection. Mattes et al. showed significant association between the detection of owl’s eye inclusion bodies and positive CMV polymerase chain reaction, and hence they concluded that histological detection of owl’s eye inclusion bodies is an insensitive but highly specific method of detecting CMV organ involvement.[6]

In our case of arteriovenous hemangioma, an incidental association or causation by CMV is not clear. Such an association has been reported in Juvenile hemangiomas.[3,4]

Conclusion

Hemangiomas of the parotid gland are frequent and the most common tumor in children. The association with CMV in non-immunocompromised living infants is extremely rare in resected specimens of salivary gland hemangioma.

References