



Submandibular hemangioma with multiple phleboliths mimicking sialolithiasis: the first pediatric case

Submandibiüler bezin sialoliti taklit eden multipl flebolitler içeren hemanjiyomu: İlk pediatrik olgu

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Hemangiomas are the most common masses of the major salivary glands in parotid glands in childhood particularly. They occur more frequently in the parotid gland and rarely the submandibular gland. Changes in blood flow dynamics within hemangiomas may induce thrombus formation and phleboliths. Cavernous hemangioma may lead to thrombophlebitis in major salivary glands in adults. To our knowledge, cavernous hemangioma of submandibular glands containing phleboliths in childhood has not been described so far in the literature. In this article, we report the first pediatric case of a cavernous hemangioma containing multiple phleboliths in the submandibular gland mimicking submandibular sialolithiasis in a seven-year-old boy.

Key Words: Hemangioma; pediatric sialolithiasis; phlebolith; submandibular gland.

Hemanjiyomlar, çocukluk çağında majör tükürük bezi, özellikle parotis bezlerinin, en sık görülen kitleleridir. Sıklıkla parotis bezinde nadiren de submandibiüler bezde görülürler. Hemanjiyom içerisinde kan akımındaki değişiklikler, trombüs oluşumu ve flebolitleri tetikleyebilir. Kavernöz hemanjiyom, erişkinlerde majör tükürük bezlerinde tromboflebite neden olabilir. Bildiğimiz kadarıyla bugüne kadar literatürde çocukluk çağında submandibiüler bezde flebolitler içeren bir kavernöz hemanjiyom olgusu tanımlanmamıştır. Bu yazıda, yedi yaşında bir erkek çocukta submandibiüler sialoliti taklit eden, çok sayıda flebolit içeren ilk pediatrik kavernöz hemanjiyom olgusu sunuldu.

Anahtar Sözcükler: Hemanjiyom; pediatrik sialolit; flebolit; submandibiüler bez.

Benign mesenchymal tumors occur more frequently in parotid glands (~85%) than submandibular glands (~10%).^[1,2] Hemangiomas are responsible for 30% of masses of salivary glands in all age groups and 60% of masses

of salivary glands in childhood.^[3] Changes in blood flow dynamics within hemangiomas result in thrombus formation and phleboliths and can lead to thrombophlebitis in major salivary glands in adults.^[1,2,4] To our knowledge, cavernous



hemangioma of submandibular glands containing phleboliths in childhood has not been described so far in the literature.

We present what we believe to be the first case of a cavernous hemangioma containing multiple phleboliths and formation phases of thrombophlebitis in submandibular gland mimicking submandibular sialolithiasis in a child.

CASE REPORT

A seven-year-old boy visited the otolaryngology service complaining of painless and slow-growing submandibular area swelling in his right neck. The swelling was not exacerbated during eating. Clinical examination revealed a painless and soft mass in the right submandibular region. Salivary flow was normal in the right submandibular gland compared with the left. The remainder of the examination was unremarkable. Computed tomography (CT) scan demonstrated an enlarged right submandibular gland including heterogeneous hypodense areas and calcified foci. Although a few sialoliths (4-8 mm in diameter) were seen, there was no duct dilation. Other major salivary glands had a normal view (Figure 1).

The submandibular gland was surgically removed via an external approach. There was an unusual bleeding during dissection from hemangiomatous areas. The hemangioma was in the salivary gland structures (Figure 2). Histopathologically, the submandibular gland tissues were normal. The lesion showed features of cavernous hemangioma such as large irregular spaces lined by endothelial cells. The vascular spaces were filled with blood cells. Additionally,

formation stages of phleboliths and calcified thrombi were observed in the hemangioma (Figures 3, 4). The lesion was diagnosed as cavernous hemangioma containing multiple phleboliths of submandibular gland. During the 25-month-follow-up, the functional and cosmetic results were excellent.

DISCUSSION

Hemangiomas are benign vascular lesions and within the first three decades they usually cause symptoms such as deformity, mass, pain and discoloration. In our case, there was only swelling in right submandibular area.

Hemangiomas are the most common salivary gland masses in childhood.^[3] They occur predominantly in the parotid gland, but they are rarely reported in the submandibular and sublingual glands.^[1,2] Most cases of submandibular hemangiomas have been reported in adults.^[1,4] Changes in blood flow dynamics within hemangiomas result in thrombus formation and phleboliths and can lead to thrombophlebitis in major salivary glands in adults.^[1,2,4] Cavernous hemangioma containing multiple phleboliths of the submandibular gland is rarely seen. So far, all reported cases are in adults.^[4-6] To the best of our knowledge, this is the first pediatric case reported in the literature.

During surgery, the lesion had an appearance different from normal gland tissue and similar to hemangioma. It was a hemorrhagic structure including lacunar spaces and calcified foci. Nevertheless, definitive diagnosis of cavernous hemangioma containing multiple phleboliths was made by pathologic examination.

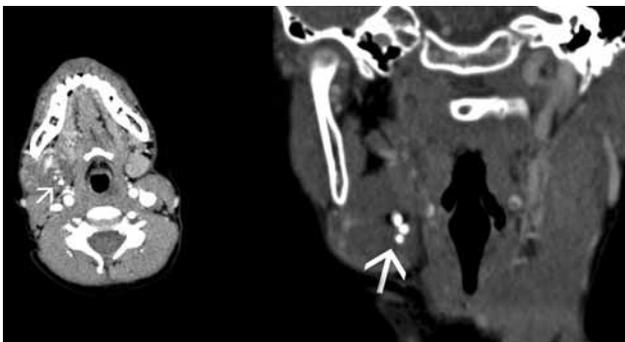


Figure 1. Right submandibular gland has heterogeneous hypodense appearance including hyperdense foci of about 8 mm diameter are seen on computed tomography.

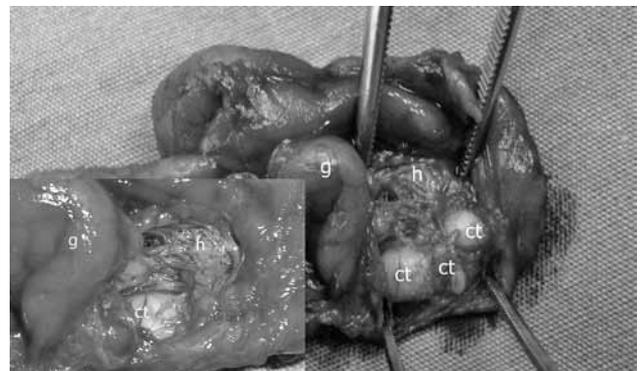


Figure 2. Hemangioma and multiple thrombophlebitis within submandibular gland tissues. h: Hemangioma; g: Gland tissue; ct: Calcified thrombophlebitis.

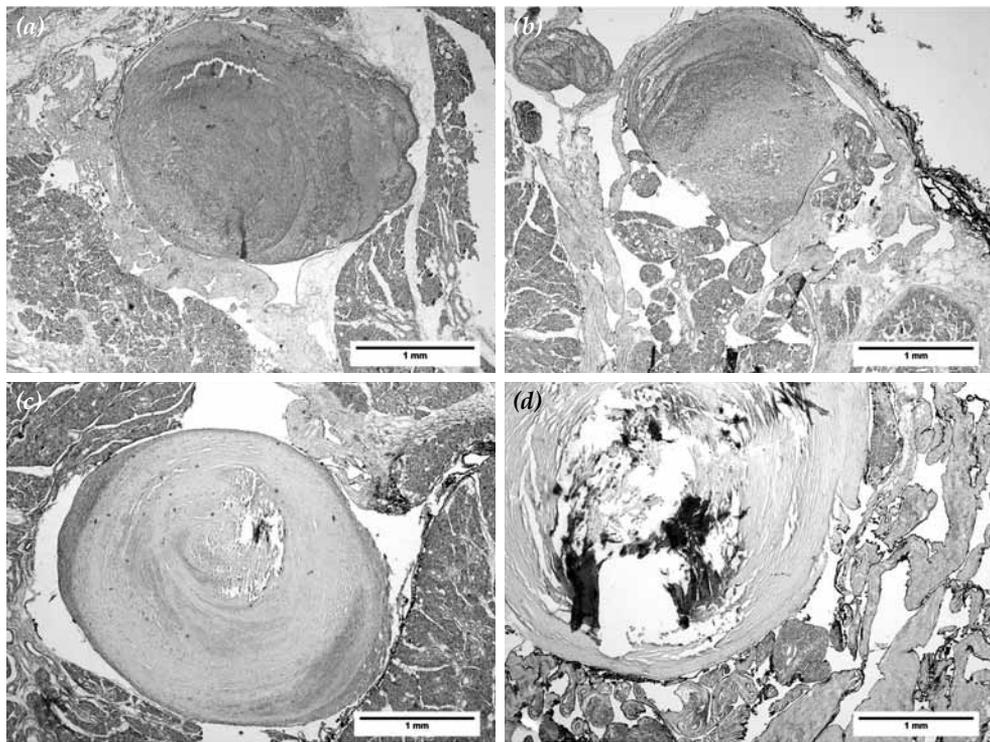


Figure 3. Formation of calcified thrombophlebitis (H-E x 40). (a) Early stage erythrocyte accumulation and fibrin deposition. (b) Organization and thrombus formation. (c) Fibrosis and calcification. (d) Common and mature calcification.

Histopathological stages and formation of calcified phleboliths (fibrin deposition and erythrocyte accumulation caused by stasis, thrombus formation, fibrosis, calcification respectively) within vascular spaces were seen by pathologic examination (Figures 3, 4).

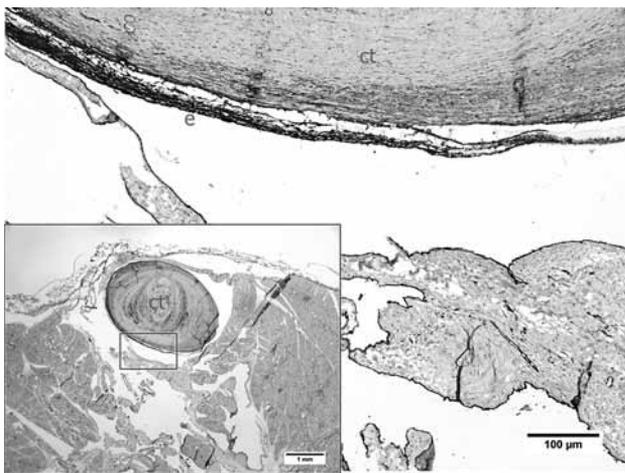


Figure 4. Endothelium stained with CD31 as brown color and calcified thrombophlebitis appearance in vascular endothelium: ct: Calcified thrombophlebitis, e: Endothelium (CD-31, 20x and 200x).

In conclusion, the present case makes us think that phleboliths can develop unexpectedly earlier than the predicted age, and may manifest in childhood. In addition, when a submandibular gland mass with heterogeneous appearance including multiple sialoliths is seen on neck CT in children, a hemangioma with multiple phleboliths should be born in mind with respect to differential diagnosis and the nature of the planned operation.

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