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Case report of a large hemangioma of the parotid gland with multiple phleboliths and tonsilloliths

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Abstract

Hemangiomas are benign vascular deformities characterized by an increased proliferation and turnover of endothelial cells. They account for the majority of parotid gland tumors in infants but are rare in adults. Changes in blood flow dynamics within hemangiomas can cause stasis, thrombus formation and phleboliths. Tonsilloliths are calcifications occurring primarily within the palatine tonsillar crypts. We report the case of a large hemangioma of the parotid gland with multiple phleboliths and tonsilloliths in an adult, highlighting the clini cal and imaging features on panoramic radiography, Cone Beam Computed Tomography (CBCT), cervical ultrasound, Magnetic Resonance Imaging (MRI) and Doppler imaging. A 20-year-old woman presented, complaining of a painless swelling below her right ear. Panoramic radiograph showed multiple randomly distributed round-to-oval radiopaque structures overlying the right mandibular ramus. CBCT revealed several radiopaque structures in the right palatine tonsillar crypts. Cervical ultrasound exposed a large heterogeneous and predominantly hypoechogenic mass in the right parotid gland. MRI displayed a well-defined lesion in the right parotid gland extending into the parapharyngeal pre-styloid space, hypointense on T1 and hyperintense on T2, containing several nodules. The diagnosis was: large hemangioma of the right parotid gland extending into the parapharyngeal pre-styloid space, with multiple phleboliths and tonsilloliths. Propranolol was delivered, with periodic follow-up on Doppler images, showing a hemangioma size reduction. Standard radiographs can detect tonsilloliths and phleboliths but additional imaging modalities disclose the exact diagnosis and location of calcifications and the diagnosis, structure and extent of the vascular lesion. Tonsilloliths and phleboliths should be considered in the differential diagnosis of radiopaque masses involving the mandibular ramus. Hemangioma with phleboliths should be considered in the differential diagnosis of parotid tumors when numerous intraglandular calcification nodules are detected on radiographs.

Keywords: Hemangioma, Phlebolith, Tonsillolith, Parotid gland, Head and Neck.

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INTRODUCTION

Hemangiomas are benign vascular tumors characterized by an increased renewal and proliferation of endothelial cells. They appear congenitally or shortly after birth, but are usually detected by the second or third decade of life and are more frequent in females [1-4]. The head and neck region hosts 65% of hemangiomas that mainly affect the salivary glands, with the parotid as the most common location (81-85% of the cases). In infants, hemangiomas account for the majority of parotid gland tumors [3,5-7]. In adults, hemangiomas of the parotid gland are relatively uncommon, with only few cases reported in the literature [8]. Histopathologically, three types of hemangiomas are described: capillary, cavernous and mixed [2]. Adult salivary gland hemangiomas are cavernous, while infantile hemangiomas are usually capillary. Cavernous hemangiomas are larger and less well circumscribed than capillary and juvenile hemangiomas [9]. Clinically, hemangiomas often have a bluish tint; pain, swelling, compressibility, pulsation, bruits, or thrills are rarely experienced [10].

Injury to a vessel wall or blood flow stagnation lead to changes in blood flow dynamics within hemangiomas, which can cause stasis, thrombus formation and phleboliths. Phleboliths are characteristic features of hemangiomas, defined as calcified nodules or thrombi, containing calcium carbonate and calcium phosphate salts [13-15]. They are multiple, randomly distributed, asymptomatic and have a variable size [10,13].

Phleboliths are identified during routine imaging on standard radiographs as oval or round radiopaque structures with concentric radiolucent or radiopaque laminations [15,16,22]. Other imaging modalities, like

Computed tomography (CT) scan, Magnetic Resonance Imaging (MRI), ultrasound and Doppler imaging contribute significantly in the diagnosis and treatment of hemangiomas. A clinical examination and imaging by CT scan or MRI determine the extent of the vascular lesion [17-21]. A biopsy and microscopic examination can help in the diagnosis [23,24].

Tonsilloliths are calcifications occurring primarily within the palatine tonsillar crypts due to chronic and recurrent inflammation. Clinically, they appear as white or yellow plaques, as hard as stone when they have existed for a long time. They may be single or multiple, unilateral or bilateral. The smaller tonsilloliths are usually multiple and asymptomatic whereas larger ones are generally solitary and may present several symptoms like irritation, pain, discomfort, foreign body sensation, swallowing and eating difficulties, halitosis, otalgia, and ulceration [28-34]. Because tonsilloliths can be asymptomatic, they are usually incidentally discovered during routine panoramic radiographs, where they appear as single or multiple radiopacities over the middle portion of the mandibular ramus [31,34,35]. However, based on the panoramic radiograph alone, it is difficult to distinguish tonsilloliths from other soft tissue calcifications in this region, like foreign bodies, odontomas, osteomas, sialoliths, phleboliths, cysticercosis, calcified lymph nodes, carotid arteriosclerosis, stylohyoid ligament calcification, idiopathic osteosclerosis, long hamular process, or osteoma cutis [33,36,37]. Therefore, CT scan was reported as the most convenient and accurate imaging modality of tonsilloliths since it provides a three-dimensional image. However, recently, CBCT was preferred over CT because it offers advantages of a significantly lower radiation exposure, shorter scanning times, higher spatial resolution, more compact design and reasonable price. On a CBCT, palatine tonsilloliths appear as radiopacities in the medial part of the mandibular ramus and the lateral wall of the oropharyngeal airway [33,34,38].

The present report describes the case of a 20-year-old woman having a large hemangioma of the parotid gland extending into the parapharyngeal pre-styloid space, associated with multiple phleboliths and tonsilloliths, highlighting the clinical and imaging features on panoramic radiography, CBCT, cervical ultrasound, MRI and Doppler imaging.

CASE REPORT

A 20-year-old woman presented to our clinic for a check up, complaining of a painless swelling below her right ear. Extraoral examination showed a diffuse soft tissue swelling in the right parotid region, causing slight facial asymmetry. A soft, elastic, painless, non-fluctuating and non-pulsatile mass was palpated at the right parotid gland. Intraoral examination revealed normal-colored mucosa and normal clear saliva discharge from the right parotid duct. In addition, multiple small white concretions were incidentally observed at the back of the throat within the right tonsillar crypts, diagnosed as tonsilloliths.

The panoramic radiograph exposed several randomly distributed round-to-oval radiopaque structures with a variable size, overlying the right mandibular ramus (Figure 1). They were suspected to be phleboliths, tonsilloliths or sialoliths. Therefore, additional imaging modalities [CBCT, cervical ultrasound, MRI and Doppler imaging (for follow-up)] were requested to determine the exact diagnosis and set a proper treatment plan.

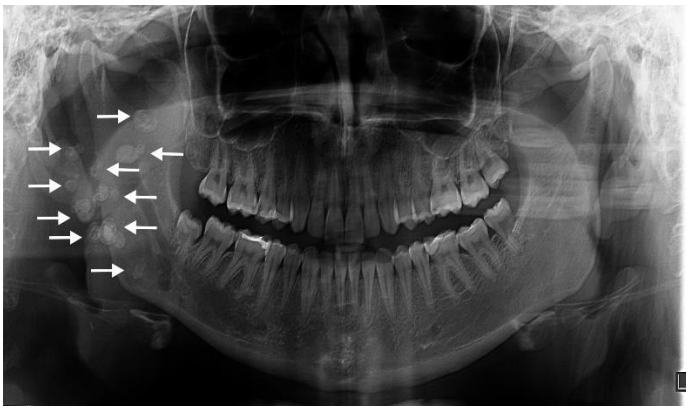

Figure 1: Panoramic radiograph exposing multiple randomly distributed round-to-oval radiopaque structures with a variable size, overlying the right mandibular ramus (white arrows).

Axial, coronal and sagittal CBCT images showed various radiopaque structures at the right palatine tonsillar crypts level corresponding to tonsilloliths (Figure 2).

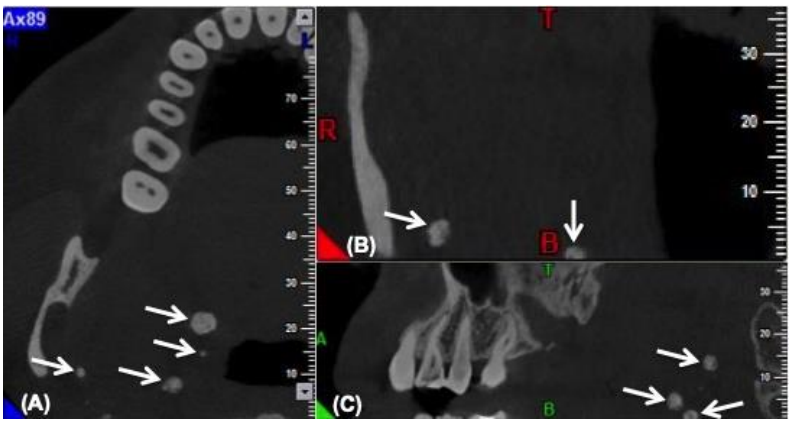

Figure 2: Axial **(A)**, Coronal **(B)** and Sagittal **(C)** CBCT images showing multiple radiopaque structures at the right palatine tonsillar crypts level corresponding to tonsilloliths (white arrows).

The cervical ultrasound revealed a large (40*17mm in axial dimensions) heterogeneous and predominantly hypoechogenic mass containing necrotic cysts in the right parotid gland. The mass is associated with some latero-cervical lymphadenopathies. An MRI was indicated to determine its nature.

The MRI exposed a process on the right side, made up of two components: a first component measuring 35*30mm in axial dimensions, located in the right parotid gland and a second more deep component measuring 45*30mm in axial dimensions developed in the parapharyngeal pre-styloid space. This process is a well-defined heterogenous lesion (hypointense on T1, hyperintense on T2), containing multiple cystic components with vascular content and several

nodules corresponding to phleboliths. Gadolinium contrast was administered, then axial T1-weighted and coronal fat-saturated T1-weighted MR images were repeated, which showed an intense and homogeneous enhancement. Fat saturation was applied on axial and coronal T2-weighted MR images and coronal T1-weighted with contrast MR image, leading to suppression of the signal from normal adipose tissue, which reduced the chemical shift artefact, improved visualization of uptake of contrast material and tissue characterization, making the lesion easier to detect (Figures 3,4).

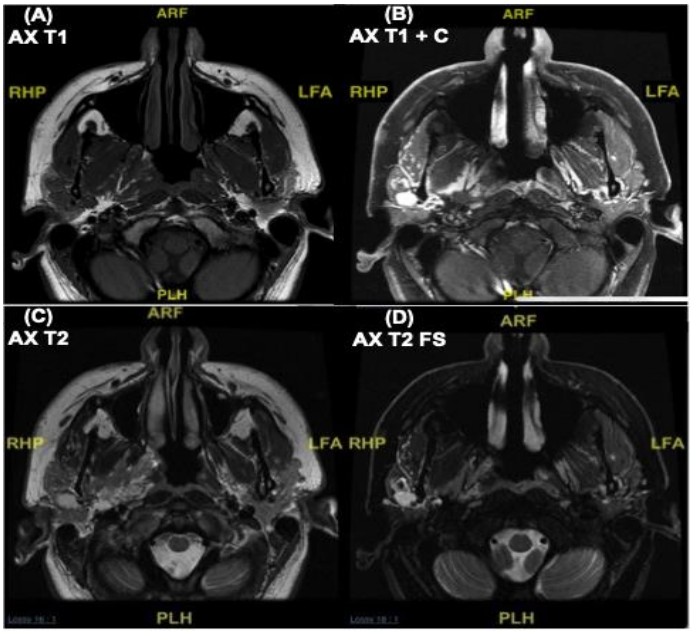

Figure 3: Axial MRI cuts at the middle of the first component of the lesion, located in the right parotid gland. **(A)** Axial T1-weighted MR image revealing a well-defined hypointense lesion that contains multiple cystic components with vascular content and several nodules corresponding to phleboliths. **(B)** Axial T1-weighted MR image with contrast showing the same image as (A) but with intense enhancement of the tumor. **(C)** Axial T2-weighted MR image displaying the same image as (A) but the tumor is hyperintense. **(D)** Axial fat-saturated T2-weighted MR image exposing the same image as (C) but the tumor is easier to visualize because of the fat saturation.

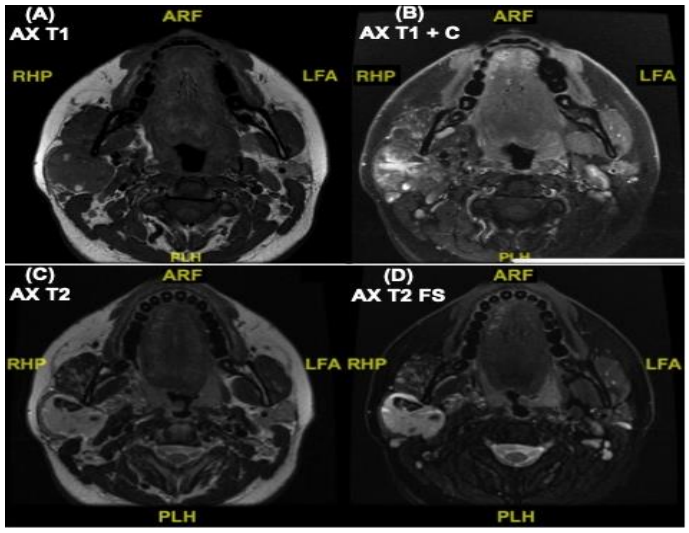

Figure 4: Axial MRI cuts at the middle of the second component of the lesion, located deeply in the parapharyngeal pre-styloid space. **(A)** Axial T1-weighted MR image revealing a well-defined hypointense lesion that contains multiple cystic components with vascular content and several nodules corresponding to phleboliths. **(B)** Axial T1-weighted MR image with contrast showing the same image as (A) but with intense enhancement of the tumor. **(C)** Axial T2-weighted MR image displaying the same image as (A) but the tumor is hyperintense. **(D)** Axial fat-saturated T2-weighted MR image exposing the same image as (C) but the tumor is easier to visualize because of the fat saturation.

According to its aspect, it was diagnosed as a benign vascular lesion, more specifically a hemangioma. Based on the findings, the diagnosis is: large hemangioma of the right parotid gland extending into the parapharyngeal pre-styloid space, associated with multiple phleboliths.

Since the tonsilloliths and phleboliths are small and asymptomatic, they were only periodically observed. The hemangioma was treated by delivering propranolol, with periodic follow-up. Follow-up Doppler images were obtained 5 and 10 months after treatment initiation. The first follow-up test showed a regression of the lesion in the right parotid gland from 35*30mm to 27*20mm in axial dimensions while the second assessment revealed a stability of the lesion size. Doppler signals were barely detected within the lesion, denoting a resistant systolic blood flow. Doppler imaging also confirmed the presence of calcifications within the lesion, corresponding to phleboliths. Following treatment, the hemangioma and swelling didn't recur. Surgical excision was not adopted because of the risk of severe hemorrhage.

DISCUSSION

Benign mesenchymal tumors affecting the major salivary glands are often difficult to diagnose, especially in the absence of symptoms and in adult patients, in whom this condition is rare and not usually considered in a differential diagnosis. If the diagnosis is only based on patient history and clinical features, the preliminary evaluation may indicate a benign mesenchymal tumor of the salivary gland or sialolithiasis [4]. 30% of benign mesenchymal tumors are hemangiomas, with the parotid gland as their most common location [40].

Head and neck phleboliths may mimic other calcifications in the same region, like sialoliths, tonsilloliths, calcified lymph nodes, foreign bodies, carotid atherosclerotic plaques, cysticercosis, healed acne lesions and miliary osteoma cutis [13]. When multiple, sialoliths appear on a single line, whereas phleboliths are randomly distributed and usually coexist with hemangiomas [15]. In such cases, a histopathological examination, clinical history, and immunohistochemistry can help in determining the final diagnosis [41]. In this case, based on clinical and panoramic radiography findings, the multiple randomly distributed round-to-oval radiopaque structures overlying the right mandibular ramus were suspected to be phleboliths, tonsilloliths or sialoliths. In fact, hemangioma with phlebolith within the salivary gland is often mistaken for sialolithiasis on x-rays, because sialolithiasis is more frequent [5,13]. But because of disadvantages of the panoramic radiography (two-dimensional image, superimposition, distortion, and magnification), the precise diagnosis can't be determined [34,36]. Therefore, additional three-dimensional imaging modalities (CT scan or CBCT) are needed. Sialography may be useful in assessing intraglandular radiopaque lesions to rule out sialoliths as a possible diagnosis. An intraoral transducer may be efficient to diagnose tonsilloliths [42]. In this case, a sialolithiasis was ruled out because of the random distribution and multiplicity of the radiopaque structures, the absence of any history or presence of a parotitis or obstructive parotid swelling or pain, and the salivary return was adequate and clear.

The diagnosis and extent of the hemangioma can only be determined by a clinical examination and imaging by CT scan or MRI but its detectability on MR images is slightly superior to that on CT images. Phleboliths detectability on CT images is superior to that on MR images [12,18,19,21]. Hemangiomas are usually perceived as masses with well-defined phleboliths on the CT but the latter can fail to properly define soft tissue planes and may not be diagnostic if phleboliths are absent [43]. On the MRI, hemangiomas are usually heterogenous, showing hyperintensity on T2-weighted images and isointensity or hypointensity

on T1-weighted images, whereas phleboliths appear as nonenhancement nodular structures in contrast-enhanced T1-weighted MR images and hypointense nodular structures in T2-weighted MR images [44,45]. Injection of Gadolinium contrast medium enhances and improves the clarity of MR images. Fat saturation suppresses the signal from normal adipose tissue, making the lesion easier to detect. On ultrasonography, hemangiomas appear as heterogeneous and predominantly hypoechogenic masses while phleboliths look as multiple hyperechoic areas within the mass [15]. Doppler ultrasonography applies high-frequency sound waves to confirm the presence of calcifications, corresponding to phleboliths [17,46,47].

In this report, we requested a panoramic radiograph, cervical ultrasound, MRI and Doppler imaging (for follow-up). A CBCT was also obtained rather than a CT scan, to have a detailed three-dimensional examination of the site and extent of the calcified structures overlying the right mandibular ramus, because CBCT has advantages over CT (mentioned in the Background) [38,48,49]. To the best of our knowledge, only one previous report [39] evaluated the mandibular ramus calcifications using CBCT imaging.

Regarding treatment, small tonsilloliths and phleboliths are harmless; if they don't bother the person (which is our patient's case), only periodic observation should be applied [50]. The therapeutic approach of hemangiomas depends on clinical factors: tumor site, extent/depth of invasion, growth rate, accessibility, patient's age and esthetic considerations. Treatment methods include using corticosteroids, propranolol, vincristine, interferon alfa-2a or 2b, cryotherapy, vascular ligation, embolization, sclerotherapy, fibrous agents and laser surgery [2,10,12,21,25,26,27]. If indicated, surgical excision is the basis of treatment. Periodic observation is also an alternative option [17]. In this case, propranolol was delivered, with periodic follow-up on Doppler images, to reduce the hemangioma size and decrease the amount of blood flowing through the vessels by constricting them, which makes the hemangioma softer and less red [51]. The first Doppler follow-up test showed a regression of the lesion size, while the second assessment revealed a stability of the lesion size.

CONCLUSION

In conclusion, this case leads to three contributions:

1. It's one of the few existing reports of a large hemangioma with multiple phleboliths of the parotid gland.

2. It's the second report evaluating the mandibular ramus calcifications using CBCT imaging.

3. It's the first report including panoramic radiography, CBCT, cervical ultrasound, MRI and Doppler images of a large hemangioma of the parotid gland with several phleboliths and tonsilloliths.

In addition, this report confirms that standard radiographs can detect tonsilloliths and phleboliths but additional imaging modalities disclose the exact diagnosis and location of calcifications and the diagnosis, structure and extent of the vascular lesion.

Moreover, this case emphasizes that tonsilloliths and phleboliths should be considered in the differential diagnosis of radiopaque masses involving the mandibular ramus. It highlights also that a hemangioma with phleboliths should be included in the differential diagnosis of a swelling in the head and neck area. More specifically regarding the parotid gland, when numerous intraglandular calcification nodules are detected in imaging studies, a hemangioma with phleboliths should be considered in the differential diagnosis of parotid tumors.

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Nil.

Conflict of Interest

The authors declare no conflict of interest.

Compliance with ethical standards

Ethics approval and consent to participate: Not applicable.

Consent for publication: Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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