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Articles in this Collection

Hypervascular Soft Tissue Tumor of the Skull Suspicious for Hemangiopericytoma: A Case Study

Joanna Hall, AAS, RDMS, RVT

Abstract
Hemangiopericytoma (HPC) is a rare mesenchymal tumor that is predominantly found in the pelvis, retroperitoneum, and lower extremities, occasionally occurring in the larynx, spleen, bones of the thorax, or the meninges. A case is presented of a hypervascular soft tissue mass of the skull suggesting a diagnosis of hemangiopericytoma. Imaging modalities including computed tomography and sonography were used to help characterize this soft tissue lesion of the skull to aid in the proper course of diagnosis and treatment. The characteristics of hemangiopericytoma are described, and note is made that because of the vascular nature of these lesions, surgical intervention is used as a final resort when there are multiple reoccurrences following radiation therapy.

Keywords
sonography, hemangiopericytoma, intracranial, meningeal, computed tomography, meningioma

Introduction
Hemangiopericytoma (HPC) is a rare mesenchymal tumor that is predominantly found in the pelvis, retroperitoneum, and lower extremities, occasionally occurring in the larynx, spleen, bones of the thorax, or the meninges.1–8 Intracranial meningeal hemangiopericytoma (M-HPC) represents a rare and aggressive intracranial neoplasm located along the dural sinuses. It constitutes less than 1% of all intracranial tumors and approximately 2% to 4% of all meningeal tumors.9,10 Clinically, these tumors can present as palpable non-tender masses when they extend into soft tissue. Many authors have argued about the true origin of this tumor. Because these tumors are considered vascular in nature, sonography is a useful tool in adjunct with other imaging modalities such as computed tomography (CT).11 Sonography is particularly able to characterize the lesion’s vascular and compositional elements. This case report describes a superficial lesion located on the left frontal/parietal portion of the scalp suspicious for hemangiopericytoma that was evaluated using both sonography and computed tomography.

Case Report
A female patient in her late 70s presented to the Radiology Department with a complaint of a “knot” on her head. Her past medical history included breast cancer. Her physical examination confirmed a non-tender palpable mass located on the left frontal/parietal portion of her scalp. The patient was referred for a soft tissue sonogram of the region of interest and was later referred for a correlative CT for the same area.

Sonographic Findings
Sonographic findings included a large hypervascular solid appearing lesion correlating to the non-tender palpable mass on the patient’s scalp. The mass measured up to 6 cm in its longitudinal dimension (cranio-caudal) and 3.3 cm in its antero-posterior dimension, obtained in the sagittal plane (Figures 1 and 2). In the transverse plane, the mass measured up to 3.3 cm in its widest dimension (Figure 3). The mass appeared to extend through the adjacent calvarium, with the bony defect measuring 2.7 cm at its widest point (Figure 3). This figure also shows small spicules of bone that were seen as multiple small echogenic foci near the defect of the calvarium, which was suggestive of a malignancy arising from the skull. The mass also appeared to extend intracranially but was not able to...
be completely characterized on this superficial sono-
graphic examination. The vascularity of the mass,
extending through the defect of the calvarium, was
shown by color Doppler imaging (Figure 4). Further
testing using CT without contrast was recommended
and obtained three weeks later.

**Computed Tomography Findings**

Computed tomography without contrast was done using 5
mm axial cuts obtained from the vertex through the base
of the skull per routine protocol (Figures 5A–5D). A
large, partially pedunculated soft tissue mass arising from
the vertex scalp of the left parietal scalp was shown. This
mass measured up to 7 cm in largest transaxial diameter.
Associated permeative destruction of the underlying pari-
etal and frontal calvaria was also shown without signifi-
cant change since the prior sonographic study. The
paranasal sinuses were clear.

No further clinical follow-up or tissue biopsy diagno-
sis was available for this patient, making the diagnosis
presumptive for hemangiopericytoma but not proven.

**Discussion**

Hemangiopericytoma is a highly vascular tumor that occurs
around capillaries and postcapillary venules. It originates
in a specific cell type called pericytes, identified by Rouget
in 1873 and subsequently described by Zimmermann in
1923. Just slightly more than 300 cases of HPC have
been reported since Stout and Murray described HPCs as
“vascular tumors arising from Zimmermann’s pericytes” in
1942. They account for 3% to 5% of soft tissue sarcomas
and only 1% of all vascular tumors with the frequency of
occurrence in the head and neck at about 16% to 33%. The
rarity of HPC and its controversial histological classifica-
tion results in its frequent misdiagnosis, thus making the
treatment quite challenging.
The World Health Organization classification of central nervous system tumors previously considered HPC tumors a very close comparison with meningiomas, but the current classification system introduced in 2007 distinguishes HPC as a separate entity. Recent studies have suggested that the histomorphology and immunophenotype of HPCs are different from that of meningioma. Presently HPCs are classified into a group of “mesenchymal, non-meningothelial tumors.” Formerly considered one of the variants of meningioma, and often referred to as “angioblastic meningioma” because these tumors are considered vascular in nature, sonography is a useful tool in adjunct with other imaging modalities such as CT. Typical of these tumors, the case reported previously was first demonstrated as a non-tender palpable soft tissue mass presenting on the patient’s skull. The hypervascular characteristics of the mass were also demonstrated using color and spectral Doppler imaging techniques with duplex sonography. Although the official final report did not state a definitive site of origin for the mass, HPCs have been noted to arise often from dural sinuses, the skull base, tentorium, and the falx cerebri. Based on the presentation of the mass demonstrated on both sonography and CT, it was believed likely that the mass was skull-based in origin.

These very aggressive tumors are characteristically highly cellular and richly vascular, tending to recur even after macroscopic total resection. A microscopic diagnosis is based on the recognition of an architecture characterized by a pericytomatous pattern. However, the same pattern occurs in a variety of neoplasms, such as fibrous histiocytoma, solitary fibrous tumor, and fibrous meningioma. Therefore, distinguishing HPC from other tumors can be difficult, especially when the characteristic features of other neoplasms are inconspicuous. However, because of differences in the prognosis and patient management, a proper diagnosis is critical. Unfortunately, there was no available follow-up information for this case.
to report such as histological results or treatment. Several authors, however, have reported that lytic destruction of the adjacent skull on plain X-rays or CT suggests an HPC while hyperostosis, or the excessive growth or thickening of bone tissue, suggests a meningoiam. Marked lytic destruction of the calvaria was demonstrated on both sonographic and CT images obtained for this case. Sonographic images demonstrated both a defect in the calvaria and spicules of bone nearby, leading to the high probability that the mass had lytic properties. Both the gyrus and sulcus of the patient’s brain tissue could also be appreciated deep to the defect of the calvaria, again suggesting that the mass was an HPC and not a meningoiam.

Meningeal HPCs can recur locally or distantly in the neural axis or as extraneural distant metastases. They have a relentless tendency for local recurrence and metastases outside the central nervous system, which can appear even many years after diagnosis and adequate treatment of the primary tumor. The local recurrence rate varies from 45% to 91% among different studies, and a 15-year distant metastasis rate approaches 70% after surgery alone. The patient in this case, however, already had a previous history of breast cancer. In a study of 21 patients carried out over a 34-year period, patients with surgical removal of an intracranial meningeal hemangiopericytoma followed by external radiotherapy had a reduced risk of local recurrence. Considering the aggressive nature of HPCs, gross total resection of the tumor is the optimal treatment method, though complete resection can be difficult or impossible because of the highly vascular nature of the lesion and the risk of bleeding tendency in the operative field. Because of this, some authors have suggested the use of radiotherapy as a neoadjuvant treatment based on the possibility that the proliferating capillaries will be obliterated after radiation. Stereotactic radiosurgery may be an alternative option for the treatment of HPCs that are less than 2 cm in diameter with radiation doses of 15 Gy or higher at the 50% isodose line.

Conclusion

The rarity of hemangiopericytomas and their unique histological properties make the correct diagnosis of such a lesion difficult based solely on diagnostic imaging findings. Some characteristics, however, including the high vascularity and the lytic properties of the mass, can be appreciated sonographically and be suggestive for HPC. Because of the aggressive and vascular nature of this lesion, it is imperative that proper technique is used to adequately represent the characteristics of the lesion so that appropriate management and treatment options can be undertaken. Sonography can be as a significant adjunct tool in assessing both the lytic and vascular components in lesions that present as soft tissue palpable masses in asymptomatic patients. This case report is meant to help sonographers characterize and distinguish the features of hypervascular tumors in order to reach a proper diagnosis.

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References

Intraductal Papilloma of the Extraglandular Segment of the Stensen Duct: A Sonographic Diagnosis and Pathological Correlation

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Abstract

Intraductal papillary neoplasm of major salivary glands is exceedingly rare. Most reported cases have been diagnosed on histopathology; an extensive literature search found no case where a preoperative imaging diagnosis was suspected or established. This report depicts the sonographic diagnosis of intraductal papilloma of the extraglandular segment of the Stensen duct. The sonographic and histological appearances of this rare entity are illustrated.

Keywords

intraductal papilloma parotid, major salivary gland papilloma, intraductal neoplasm salivary

Tumors arising from the major ducts of salivary glands are distinctly rare, with less than 150 reported cases.¹ They are most commonly encountered in minor salivary glands. Involvement of major salivary glands is even more rare, with those arising from the extraglandular segment of the duct exceedingly rare.² Most reported cases are benign, and malignant transformation is uncommon, with only two reported cases.³

Unlike their counterparts in the breast, where extensive literature is available on the imaging features, little literature is available on the imaging features of intraductal salivary gland tumors. Most of the available reports are from pathology, and a comprehensive literature search has revealed no reported cases where a diagnosis of salivary gland ductal papilloma was demonstrated by preoperative sonographic imaging.³

This case reports a case of extraglandular Stensen duct papilloma with focal malignant change that was diagnosed on sonography and confirmed on histopathology. The unique features of our case are its involvement of the parotid salivary gland, location in the extraglandular segment of the Stensen duct, preoperative diagnosis, and malignant change in the papilloma.

Case Report

A woman in her mid-20s presented to the otolaryngology outpatient department with a swelling over the left cheek. She also complained of occasional blood-stained saliva. On examination, a firm 3 × 1.5-cm nodule was palpable over the left cheek that was mobile over the underlying muscle and was nontender. Bloody discharge was noted through the intraoral orifice of the parotid duct. High-resolution sonography was carried out on Philips HDI 5000 machine (Philips Healthcare, Andover, Massachusetts) using a linear 7- to 12-MHz probe that revealed a dilated Stensen duct measuring approximately 1 cm in diameter. An intraductal lobulated soft tissue lesion was seen filling the proximal portion of the Stensen duct, measuring about 2.5 cm in length (Figure 1). The lesion revealed internal vascularity on Doppler examination (Figure 2). Duct walls were well visualized with no evidence of extension of the lesion outside the duct. The ipsilateral parotid gland was atrophic with no focal lesion (Figure 3). A magnetic resonance imaging (MRI) examination of the parotid gland

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revealed a dilated Stensen duct that was hypointense on T1-weighted images and hyperintense on T2-weighted images (Figure 4). However, no obvious intraductal soft tissue mass could be appreciated. An imaging diagnosis of intraductal neoplasm was made.

Upon surgical examination, a tumor was detected in the proximal part of the Stensen duct, which was dilated. It was excised with the adjacent parotid gland. On histopathology, the tumor was seen to occupy the distended lumen of the duct. It had complex papillary architecture with anastomotic columns of epithelial cells (Figure 5). These cells were moderately sized, had centrally located nuclei, and exhibited minimal pleomorphism. Occasional cells had cytoplasmic vacuoles that were confirmed by special stains to be mucinous in nature. In one focus, the tumor cells were seen in the absence of a basement membrane, a feature highly suggestive of microinvasion. There was histological evidence of acinar atrophy and chronic sialadenitis consistent with obstruction. A final diagnosis of intraductal papilloma with focal malignant change was made. The patient had an uneventful postoperative period and was apparently well at one-year follow-up with no recurrence of the tumor.

Discussion

Although many tumors arise in salivary glands, those originating in the major ducts are extremely rare. The term ductal papilloma is used to identify a group of three rare benign tumors originating from the major ducts sharing a papillary architecture. They are inverted ductal papilloma, sialadenoma papilliferum, and intraductal papilloma.1 These tend to occur in the middle aged and elderly and are rare in children and young adults. Usually the patients present with an asymptomatic nodular swelling. However, blood-stained saliva as seen in our case may occasionally be present.1

Among these three, inverted ductal papilloma is most common. In a study of 19 cases of ductal papillomas, 13 were inverted ductal papilloma and 3 each sialadenoma papilliferum and intraductal papilloma. All these three types commonly involve minor salivary glands and are infrequently seen in major glands.1 In a literature review by Brannon et al.1 of 40 cases of intraductal papilloma of the salivary gland origin, only 9 involved major salivary glands (4 in the parotid gland, 3 in the submandibular gland, and 2 in the sublingual gland). Nagao et al.3 reported only two cases of intraductal papillomas of major salivary glands, one each in the sublingual and major duct of the
The second reported case involved malignant transformation, which, according to them, was previously not reported in literature. An extensive literature search found nothing related to the imaging features of ductal papillomas, possibly because most of the reported cases are from minor salivary glands, presenting as nodular submucosal masses where a conservative local excision was both diagnostic and therapeutic. Extensive literature, however, is available on the sonographic spectrum of intraductal papillary neoplasms of the breast. Three basic sonographic appearances have been described: intraductal mass with or without dilated duct, intracystic mass, and a predominantly solid pattern with an intraductal mass totally filling the duct. Color Doppler often shows a central vascular core with branching vessels arborising within the mass. A dilated duct with an intraductal mass is considered a hallmark of breast intraductal papilloma.

The findings in the present case were similar to those of breast papilloma as it showed a lobulated hypoechoic soft tissue mass within the dilated duct with branching intratumoral vessels. MRI detected a dilated duct and could not identify the mass, possibly because the mass had similar intensity to the duct fluid on both T1- and T2-weighted sequences. Contrast-enhanced MRI would have been helpful but could not be done because of cost constraints.

Clinical differential diagnosis of a nodular lesion arising in the soft tissue of the cheek such as in the case presented would include epidermal cyst or lymph node, neoplasms of the parotid gland, lesions of the Stensen duct, and neoplasms of the accessory parotid gland. Imaging can help in differentiating all these. Because of the superficial location, high-resolution sonography should be the primary imaging modality. Tumors of the minor salivary glands, epidermal cyst, and lymph node will be separate from the Stensen duct and will not cause its dilatation. If a dilated Stensen duct is found, the possible differentials are...
sialolithiasis, benign ductal neoplasm, ductal carcinoma, or rarely hyperplasia as a cause of duct obstruction. In sialolithiasis, an echogenic calculus with posterior acoustic shadowing will be seen with proximal duct obstruction. Benign ductal papilloma, as in the presented case, will show an intraductal soft tissue mass with vascularity. Malignant Stensen duct tumor can also show intraductal soft tissue mass; however, extraductal extension and an infiltrative growth pattern will help in differentiation.

On histopathology, ductal papillomas are adenomas with unique papillary features. Specific histopathologic features allow differentiation of the three types. Sialadenoma papilliferum shows a biphasic growth pattern with an exophytic papillary component surfaced by keratotic squamous epithelium and an endophytic adenomatous component at the base of the lesion. The epidermoid cell component of the inverted ductal papilloma is the main feature differentiating it from an intraductal papilloma. Whereas intraductal papilloma is a well-circumscribed unicystic lesion, the epidermoid component of the inverted ductal papilloma expands or pushes into the surrounding connective tissue. Inverted ductal papilloma demonstrates epithelial proliferation within the excretory duct, leading to an opening of the lesion onto the mucosal surface. Intraductal papilloma is situated in the duct at a deeper level relative to the mucosal surface.1,6

Surgical excision is the treatment of choice, and recurrence has not been reported in benign cases. Although no recurrence was seen in the two reported cases of intraductal papilloma with malignant transformation,3 a theoretical possibility remains and follow-up would be prudent.

Conclusion
This is the first known report on the preoperative imaging diagnosis of an intraductal papilloma of a major salivary gland, describing the sonographic features. The unusual location in the Stensen duct and malignant transformation further add to the exclusivity of this case.

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References
Midline Ectopic Thyroid Mimicking as a Thyroglossal Duct Cyst

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Abstract
Ectopic thyroid tissue is a rare developmental abnormality. A case is reported here of ectopic thyroid tissue that presented as a midline mass, mimicking a thyroglossal duct cyst. The thyroid gland was not visualized in its normal position. This report highlights the importance of imaging in the preoperative evaluation of a midline neck mass.

Keywords
thyroid, ectopic, midline, sonography

Introduction
Ectopic thyroid tissue is a rare developmental anomaly in which there is abnormal embryogenesis of the thyroid gland. The thyroid normally descends from the floor of the primitive foregut to reach the final position in front of the trachea in the seventh week of gestation. When the normal migration of the thyroid fails, it results in an ectopic thyroid.¹–⁴ Ectopic thyroid is most common in females,¹ and it is more common in populations of Asian origin.² The prevalence is about 1 per 100 000 to 300 000 people.³ It may occur at any age but is most common at younger ages; in the series of Gopal et al.,² the mean age at presentation was approximately 14 years. A case is reported in the following of ectopic thyroid tissue presenting as a midline neck mass in a young male mimicking a thyroglossal duct cyst.

Case Report
A 19-year-old man with a provisional diagnosis of thyroglossal duct cyst (TGDC) was referred to the radiology department for sonographic examination. The patient gave a history of a midline neck mass for the past two years. The mass was small to begin with but had increased to its present size in the last few days. There was no history of dysphagia or dyspnea. There was no history suggestive of hypothyroidism or hyperthyroidism. On examination, the general health of the patient was good. A firm midline neck mass in the region of the hyoid, measuring approximately 4 × 3 cm, was seen moving with deglutition and protrusion of the tongue (Figure 1). Routine hematological parameters were within normal limits; thyroid stimulating hormone (TSH) was noted to be elevated at 37.71 mIU/L (normal values, 0.4–5.0 microIU/ml).

Figure 1. Photograph of the patient showing a 3 × 4 cm midline neck mass in the superior aspect of the neck in the region of the hyoid.

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Sonography was done using an Esaote My Lab 60 (Esaote Ultrasound, Esaote, Genoa, Italy) with a 4 to 13 MHz broadband linear array transducer. The sonogram showed nonvisualization of the thyroid gland at its normal anatomical position (Figure 2). At the site of the midline mass, an oval-shaped lesion measuring 3.00 × 1.56 × 2.37 cm, extending from midline toward the right, with echogenicity resembling that of the thyroid was seen. Another contiguous rounded lesion was seen to be extending from the aforementioned lesion, from midline toward the left side and measuring 2.40 × 1.90 × 0.75 cm, with heterogenous echogenicity showing a honeycombing pattern (Figure 3). Sonographic image morphology was suggestive of ectopic thyroid. The patient was advised to have a Technetium-99m (99mTc) nuclear thyroid scan so that findings could be confirmed. The thyroid scan showed evidence of increased tracer uptake in the midline mass; no other thyroid tissue was
seen (Figure 4). The findings were suggestive of ectopic thyroid gland. A computed tomographic (CT) scan was done to delineate the relationship of the mass to the surrounding structures of the neck (Figure 5).

Replacement therapy with L-thyroxin, 50 microgm/day, was started, and no surgery was performed. On follow-up after 6 months, the patient was doing well. A repeat TSH measured 1.26 mIU/L, in the normal range. There was also a slight reduction in the size of the mass noted.

**Discussion**

Ectopic thyroid tissue, defined as thyroid tissue not located antero-laterally to the second and fourth tracheal cartilages, is rare. It was first described by Hickman in 1869. It is a congenital disease caused by abnormal migration of thyroid tissue in the embryonic stage. The embryonic thyroid gland travels through thyroglossal duct to reach its normal position. It may never travel and remain at the base of the tongue, resulting in a “lingual thyroid,” or the descent of the thyroid may not proceed normally, leading to a variety of possible anomalous locations of the gland. In the majority of cases, it is located in the midline, between the foramen caecum and the proper location of the thyroid.

Thyroglossal duct cyst (TGDC) is another congenital defect. When the thyroid gland forms during embryonic development, it begins at the base of the tongue and moves down the neck through a canal called the thyroglossal duct. This duct normally disappears once the thyroid reaches its final position in the neck, but sometimes portions of the duct remain. They form cysts, filled with fluid or mucus, which can become infected. Confusion can arise, and it may not be possible to differentiate an ectopic midline thyroid from a TGDC clinically. Up to 1% to 2% of patients, presenting with what appears to be a thyroglossal duct cyst, have an ectopic thyroid gland. This is the case of the patient being reported, who was clinically diagnosed as having TGDC but was a case of ectopic thyroid. Of note, TGDC and ectopic thyroid can coexist. The wall of a thyroglossal duct cyst is the second most common site for ectopic thyroid tissue, the most common site being a lingual thyroid.

Ectopic thyroid is mostly asymptomatic, but like the normal thyroid tissue, it can become goitrous, can be hyper- or hypo-functioning, and can also undergo benign and neoplastic changes. These conditions cause enlargement of the gland, resulting in obstructive symptoms. Ectopic thyroid is commonly detected during periods of increased demand for thyroid hormones such as puberty and pregnancy. Our patient also noticed the midline mass only in recent years and never gave a history of having such a mass before.

Imaging, specifically sonography and nuclear thyroid scans, is a must before planning any surgery for a midline neck mass. Sonography can identify and localize the midline mass, and it can determine the solid versus cystic nature of the mass. It also shows the presence or absence of the thyroid in its normal location. CT, if done, shows the mass in relation to other neck structures in all three dimensions. A thyroid scan using $^{99m}$Tc is the most important diagnostic tool to detect ectopic thyroid tissue and should confirm the sonographic findings. It may also highlight additional sites of thyroid tissue. It has been shown to be greater than 95% sensitive and specific for differentiating ectopic thyroid tissue from other midline neck masses.

**Conclusion**

Ectopic thyroid tissue poses difficult diagnostic and management challenges. This case report demonstrates the essential role of sonography and the nuclear thyroid scan in the evaluation of a midline neck mass to prevent unnecessary or inadvertent removal of the only functioning thyroid tissue and subsequent complications.
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Figure 5. Computed tomographic scan of the neck showing (A) transverse, (B) sagittal, and (C) coronal views of the midline mass (arrows).
The presence of calculi within the human body is not uncommon, but the presence of calculi within the salivary glands is. Sialolithiasis is the presence of calculi, or stones, in the salivary glands or ducts. This condition has a 1% incidence at autopsy, but only about 50 in every 1,000,000 present symptomatically per year. Possible signs and symptoms include an upper neck or jaw mass that enlarges and recedes following meals and xerostomia, or dry mouth. Sonography has been used along with a number of other imaging modalities to diagnose this condition. The use of real-time sonographic imaging allows for identification and confirmation of the presence of stones within the salivary gland. Treatment of sialolithiasis generally begins conservatively but can result in surgery if not resolved. This case report demonstrates the use of sonography as an effective method of diagnosing sialolithiasis in a patient with classic symptoms.

Case Report

A woman in her late 60s presented in the emergency department with a mass at the base of her left jaw. The patient reported a rapid onset of swelling that evening but remarked that it was not painful. She also noted that she had experienced slight xerostomia (dry mouth) on the left side of her mouth. After palpation of the mass for any sign of calcification, the patient was taken for a computed tomography (CT) scan.

The CT was ordered with and without contrast, but because of the patient’s allergy to iodine, the CT was performed only without contrast. A marker was placed in the left submandibular region, and the CT showed no obvious evidence of fluid collection, although the metallic marker somewhat obscured several images. Without the use of contrast, an abscess could not be ruled out. No tracheal effacement was evident, and all remaining soft tissue and bony structures were unremarkable. A large density consistent with calcification was noted near the base of the left jaw (Figure 1). Sonography was suggested for further evaluation of the mass.

Sonography was performed on the soft tissues of the neck below the left mandible using an Acuson Sequoia C512 system (Siemens Medical Solutions, Malvern, Pennsylvania) with a linear-array 14-MHz transducer. The mass was thoroughly examined in the long and transverse planes (Figures 2 and 3), including Doppler assessment of blood flow to the area (Figure 4). A heterogeneous well-circumscribed mass was visualized. The mass measured 3.8 × 3 × 1.8 cm, and diffuse, normal blood flow was demonstrated within and around the area. Cystic structures lacking the presence of flow were visualized throughout the mass (Figure 5). An area of possible calcification was noted, with posterior acoustic shadowing present (Figures 5 and 6).

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These traits were indicative of a salivary duct stone. The soft tissue mass was characteristic of submandibular salivary gland tissue. The patient was referred to an ear, nose, and throat (ENT) specialist for further evaluation.

The ENT specialist saw the patient as an inpatient in the hospital. He determined that there were stones present in the gland and diagnosed her with sialolithiasis. He noted that due to the stones, the swelling of the gland would continue to increase and decrease. He determined that no further treatment was necessary at this time. The patient was directed to stay well hydrated and suck on sugar-free lemon drops to keep the saliva moving. If the patient noted a large increase in swelling again, she was
directed to return to his office for further evaluation and treatment.

**Discussion**

There are three types of salivary glands located around the mouth: the parotid glands, the submandibular glands, and the sublingual glands. The salivary glands are responsible for producing saliva and delivering it to the mouth when stimulated. Saliva is carried to the mouth through a system of ducts. The main ducts are the Stensen duct, which drains the parotid gland, and the Wharton duct, which drains the submandibular gland and part of the sublingual gland. The Stensen duct is longer and narrower than the Wharton duct; however, saliva is able to flow with gravity into the oral cavity.

Calculi are formed due to a relative stagnation of calcium-rich saliva in a setting of partial obstruction. Stones are largely composed of calcium phosphate and calcium carbonate as well as other salts and organic material such as glycoproteins, desquamated cellular residue, and mucopolysaccharides. The submandibular gland is the most common site for calculi, with an incidence rate of 80% to 92%. Reasons for this high occurrence are due largely to the duct itself. The Wharton duct is large and long, and salivary flow is slow and against gravity. Saliva in this duct also tends to be more alkaline, with a high mucin and calcium content. Other factors that may increase the incidence of stones are the presence of dehydration, use of anticholinergic medications such as Effexor, and trauma-induced predisposition to stones. Patients who experience chronic sialadenitis, or inflammation of the salivary gland, are also at a greater risk of developing calculi. Common causes of sialadenitis include postoperative dehydration, radiation therapy, and immunosuppression such as diabetes mellitus, organ transplant, chemotherapy, and human immunodeficiency virus (HIV). The patient in this case was later noted to be under treatment for cancer and currently on hospice care, which contributed to her increased risk for developing sialolithiasis.

Formation of stones in the salivary glands is more common in men than in women, and it is rare in children. The majority of patients are between 30 and 60 years of age, and 75% present with single stones. Submandibular stones tend to be larger and intraductal, whereas parotid stones are usually smaller and multiple. Differential diagnoses associated with sialolithiasis include sialadenitis, both viral and bacterial; radiation sialadenitis; lymphoepithelial cysts secondary to HIV; Sjögren syndrome; malnutrition; and tumors. Sjögren syndrome is an immune system disorder characterized by dry mouth and eyes. In a study performed by Shimizu et al, three cases of patients with Sjögren syndrome were examined with sonography. The purpose of the examination was to evaluate the damage in the glands due to chronic xerostomia. In all three patients, microliths were found in the parenchyma of the parotid gland, most of which did not demonstrate typical characteristics of calculi such as comet tail artifact or posterior shadowing. In all three cases, an additional imaging modality was used and confirmed the presence of stones in the parenchyma of the parotid gland.

Although CT, magnetic resonance (MR) imaging, and x-ray can be used to diagnose sialolithiasis, the most effective diagnostic imaging modality is sonography. Ultrasound is noninvasive, fast, and widely available, making it a first choice when assessing for sialolithiasis. Although results are operator dependent, sonography can detect up to 90% of salivary duct stones, despite that approximately 20% to 40% of salivary duct stones are not opaque on plain films. A study conducted by Jager et al examined the possibility of MR sialography, which uses evoked salivation as a contrast as opposed to an invasive contrast substance, as an alternative imaging method. Because of its noninvasive approach, MR sialography was considered superior to CT or digital radiographic sialography. The authors concluded that the use of MR sialography in delineating the submandibular ductal system and detecting stones was similar in accuracy to digital sialography and superior to sonography due to increased specificity and sensitivity, but with added expense. On the basis of these results, MR sialography would have been a possible alternative imaging modality in this case to further confirm the presence of calculi in the salivary ducts.

Treatment of sialolithiasis usually begins conservatively with a number of measures, depending on the cause of the stones. Initial treatments include hydration, warm compresses, and massage of the gland. It is often

![Figure 6. Transverse view demonstrating area of calcification (arrow) with acoustic shadowing.](image-url)
recommended that the patient use a sialagogue, such as sugar-free lemon drops, to continue salivary gland stimulation and keep saliva moving.\textsuperscript{3} If the calculi are caused by an anticholinergic medication, it is recommended that the patient discontinue use of the medication. Nonsteroidal anti-inflammatory drugs (NSAIDs) are recommended for pain, and antibiotics may be prescribed if infection is suspected. Stones that are less than 2 mm in diameter often pass spontaneously and do not require surgery.\textsuperscript{2}

If a conservative approach is not effective, a more aggressive treatment may be pursued transorally. If a stone is near the opening of the Wharton duct, the duct may be cannulated, dilated, and the stone removed. Stones that are deeper may require surgical excision of the duct or the entire gland. Extracorporeal shock wave lithotripsy may also be used on intraductal stones that are less than 7 mm and easily located by sonography. Finally, wire basket extraction under fluoroscopic guidance may be used for stones that are extraglandular and mobile.\textsuperscript{2}

**Conclusion**

Sialolithiasis is a rare salivary gland disorder characterized by the presence of calculi within the salivary gland or duct. Although it is possible to diagnose this condition using a number of imaging modalities, the most cost-effective mode is sonography, detecting up to 90\% of salivary duct stones. Although treatment can become extensive and lead to surgery, most stones can be treated simply with hydration, warm compresses, massage, and the use of sialogogues.

This case demonstrated the use of two imaging modalities to identify and diagnose sialolithiasis: sonography and CT. Sonography was shown to be an effective way of confirming a diagnosis of stones in the salivary gland. A conservative method of treatment was used to treat the patient; however, if symptoms worsen, ductal cannulation may be considered.

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**References**

Secondary hyperparathyroidism (SHPT) is one of the most common abnormalities of mineral metabolism in patients with chronic kidney disease (CKD). It is characterized by hyperplasia of the parathyroid glands and increased plasma levels of parathyroid hormone (PTH). The resulting disturbances in vitamin D, phosphorus, calcium, and PTH metabolism lead to bone disorders and multiple cardiovascular complications that contribute to the excessive morbidity and mortality of patients with end-stage renal disease (ESRD).1 Sonography has been shown to be a valuable modality for the detection of enlarged parathyroid glands in patients with SHPT.2

Case Report
A man in his 60s with a history of ESRD secondary to focal segmental glomerulosclerosis underwent a computed tomography (CT) pulmonary embolus study for new onset of atrial flutter. Incidental findings of multiple suspicious supraclavicular lymph nodes led to a referral for a neck sonogram. Sonography of the thyroid and neck was performed using a Philips iU-22 ultrasound system (Philips Medical, Bothell, Washington) with a 5- to 12-MHz linear transducer and a 5- to 8-MHz curvilinear transducer. Bilateral hypoechoic solid nodules corresponding to the CT findings were noted posterior to each thyroid lobe (Figure 1). The right nodule measured $1.3 \times 1.2 \times 1.2$ cm, and the left nodule measured $1.8 \times 1.1 \times 1.6$ cm (Figures 2 and 3). Both nodules showed mild peripheral color Doppler flow (Figure 4) and were considered suspicious for parathyroid adenomas. Exophytic thyroid nodules and lymphadenopathy were thought to be less likely. In addition, an ill-defined slightly heterogeneous isoechoic nodule was found in the mid-portion of the right thyroid lobe. This nodule measured $1.2 \times 1.1 \times 0.9$ cm and had peripheral color Doppler flow but no other suspicious sonographic features. No abnormal central or lateral lymph nodes were noted during this examination.

Ultrasound-guided fine-needle aspiration was performed on each nodule. Biopsy confirmed the bilateral posterior nodules to contain parathyroid cells. The mid-right thyroid nodule was found to be a benign follicular nodule with focal cystic degeneration. At the time of the sonogram, the patient’s laboratory values showed high parathyroid hormone, calcium, and phosphorus serum/plasma levels, which were all consistent with a diagnosis of SHPT. The patient also had elevated serum urea nitrogen and creatinine levels and a low glomerular filtration rate (GFR). He is currently being treated with chronic hemodialysis (HD), B complex vitamin, and sevelamer carbonate (Renvela; Sanofi-Aventis, Bridgewater, New Jersey) to treat his CKD and cinacalcet (Sensipar; Amgen, Thousand Oaks, California) to reduce the PTH level.

Discussion
Parathyroid hormone is secreted by the parathyroid gland and functions to maintain ionized calcium and phosphate within a normal reference range by stimulating specific
receptor-mediated responses in cells throughout the body. If a decrease in circulating ionized calcium occurs, PTH increases, which triggers three major processes aimed at restoring a normal circulating concentration: receptor-mediated tubular reabsorption of calcium occurs in the kidneys, stimulation of osteoclast resorption to release skeletal calcium occurs in the bones, and production of vitamin D and an increase in calcium absorption occur in the bowel. Once an appropriate calcium level is achieved, PTH secretion is decreased via a classic endocrine feedback loop. A disruption of any component of this regulatory system may cause excessive secretion of PTH and hyperparathyroidism.3

Secondary hyperparathyroidism is a common complication in CKD, characterized by an increase in PTH and disorders of parathyroid cell proliferation.4 The prevalence of SHPT increases as the GFR decreases.5 Secondary hyperparathyroidism develops in CKD as an adaptive response to the lowered ionized calcium level that occurs as a result of deteriorating renal function. A combination of additive factors contributes to the increase in PTH, since GFR decreases with progressive stages of CKD. As GFR falls, phosphate is increasingly retained and hypocalcemia progresses, both of which stimulate the synthesis and secretion of PTH. The circulating vitamin D level decreases in response to a reduction in functional renal tissue.3 Parathyroid gland hyperplasia occurs as the parathyroid gland compensates for the low serum calcium levels.6 Secondary hyperparathyroidism is a significant contributing factor in metabolic bone, multiple cardiovascular complications, fractures, and the increased morbidity and mortality of patients on hemodialysis.4

Chronic renal failure is best treated with renal transplantation, which should successfully restore vitamin D metabolism and prevent the progression of SHPT.5 When renal transplantation is not an available option, alternative treatment of SHPT includes oral or intravenous administration of calcium or non–calcium-based phosphate binders to reduce blood serum levels.1

Figure 1. Transverse image of the neck showing bilateral hypoechoic masses (arrows) posterior to the thyroid gland.

Figure 2. Transverse image of the right thyroid gland showing a round hypoechoic mass (arrow) posterior to the gland.

Figure 3. Longitudinal image of the hypoechoic mass inferior to the hyoid bone and posterior to the left thyroid gland.

Figure 4. Color Doppler longitudinal image of the mass inferior to the left thyroid gland showing peripheral flow.
administration of vitamin D analogues may be used to prevent the progression of bone disease. Cinacalcet is a calcimimetic drug that is used in the treatment of CKD to reduce the secretion of PTH in patients undergoing hemodialysis or peritoneal dialysis. Typically, however, combinations of medications are needed to successfully treat patients with SHPT. In addition, two surgical options are available to treat SHPT. These include total cervical parathyroidectomy with immediate heterotopic transplantation of parathyroid tissue to a distant site, often the forearm, or a subtotal parathyroidectomy, leaving a remnant of well-vascularized parathyroid tissue in the neck.

Most individuals have two superior and two inferior parathyroid glands. The superior glands are usually located posterior to the middle or superior portion of the thyroid gland. There is more variability in the location of the inferior glands, but they are most often found inferior or just posterior to the lower pole of the thyroid gland. The average size of a normal parathyroid gland is $5 \times 3 \times 2$ mm. Thus, normal parathyroid glands are rarely visualized sonographically because of their small size and insufficient acoustic differentiation from surrounding tissue. In contrast, parathyroid adenomas and hyperplasia are usually hypoechoic because of their compact cellularity relative to thyroid tissue. Parathyroid adenomas are usually well-circumscribed solid masses that are ovoid, bilobed, triangular, or longitudinal in shape. Parathyroid adenomas generally have a similar appearance to hyperplastic glands but tend to be somewhat more echogenic. Patients with SHPT do not always have symmetric enlargement of the parathyroid glands. Sonographic evaluation of SHPT is usually accurate in showing the number and size of parathyroid glands as well as their echostructural and vascular patterns. Sonography has been found to correlate well with the severity of SHPT and is an effective means of monitoring the therapeutic outcome of patients with moderate disease.

**Conclusion**

Secondary hyperparathyroidism is a common complication in CKD. This diagnosis is important for the management and treatment of bone and cardiovascular problems that may result from SHPT and lead to early mortality. Sonography aids in the detection and diagnosis of SHPT and in the monitoring of these patients during their treatment. Sonography provides a simple and effective means to differentiate thyroid and lymph node pathology from parathyroid adenomas or hyperplasia in patients with SHPT.

**Declaration of Conflicting Interests**

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**References**

Instructions

1. All SDMS CME tests must be completed through the SDMS website at http://www.sdms.org/cme. Note that test questions online may not appear in the same order as the printed test below.
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Article: JDMS CME Collection Series: Head/Neck/Thyroid
Category: Abdominal [AB]
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1. A highly vascular tumor that occurs around capillaries and postcapillary venules is consistent with
   A. Thyroglossal duct cyst
   B. Hemangiopericytoma
   C. Intraductal papilloma
   D. Hyperparathyroidism

2. Secondary hyperparathyroidism is one of the most common abnormalities of mineral metabolism associated with
   A. Decreased plasma levels of parathyroid hormone
   B. Primary hyperparathyroidism
   C. Hemangiopericytoma
   D. Chronic kidney disease

3. Tumors arising from the major ducts of the salivary glands are
   A. Typically bilateral
   B. More common in children and adolescents
   C. Often accompanied with swelling and fever
   D. A rare finding

4. The Stensen Duct is located
   A. Adjacent to the parotid gland
   B. In the sublingual region
   C. Adjacent to the submandibular gland
   D. Inferior to the tongue

5. Tumors in the major salivary ducts are typically
   A. Accompanied with bloody discharge in the mouth
   B. Benign
   C. Reported in children and young adults
   D. Metastatic malignancies

6. A differential diagnosis to consider for dilated Stensen Duct would be
   A. Enlarged lymph node
   B. Epidermal cyst
   C. Ductal hyperplasia
   D. Tumor of an adjacent minor salivary gland

7. An echogenic structure with acoustic posterior shadowing along with proximal dilation of the Stensen Duct would be consistent with
   A. Malignant ductal papilloma
   B. Benign ductal papilloma
   C. Chronic kidney disease
   D. Sialolithiasis

8. The thyroid descends to its final position in front of the trachea
   A. In the 4th week of gestation
   B. In the 7th week of gestation
   C. In the 11th week of gestation
   D. In the 16th week of gestation

9. Ectopic thyroid would be more likely to occur in which of the following population groups?
   A. Afro-American adolescent males
   B. Caucasian adult females
   C. Asian adolescent females
   D. Hispanic adult males

10. The most important diagnostic tool to detect ectopic thyroid tissue is
    A. Manual palpation
    B. High-frequency sonography
    C. Computed Tomography
    D. Technetium-99m nuclear scan

11. The most common site for an ectopic thyroid is
    A. Lingual region
    B. Antero-lateral to the second and fourth trachea cartilages
    C. The wall of a thyroglossal duct cyst
    D. Suprasternal region

12. The formation of stones in the salivary gland is most common in
    A. Men
    B. Women
    C. Adolescents
    D. Children
13. Xerostomia is a medical term describing
A. Swelling
B. Displacement
C. Excess saliva
D. Dry mouth

14. The Wharton Duct is located
A. Adjacent to the parotid gland
B. Inferior to the parathyroid gland
C. Adjacent to the submandibular gland
D. Superior to the Stensen Duct

15. The most effective diagnostic tool to detect sialolithiasis is
A. Plain film radiography
B. Computed tomography
C. Magnetic resonance imaging
D. Sonography