Ultrasonography: A Noninvasive Tool to Diagnose a Caliber-Persistent Labial Artery, an Enlarged Artery of the Lip

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Objective. Currently, practitioners use clinical and histopathologic examination to diagnose a caliber-persistent labial artery (CPLA). We illustrate the use of ultrasonography as a noninvasive diagnostic tool to visualize this enlarged artery of the lip. ***Methods.*** We examined the lips of 3 patients with a suspected CPLA. We localized and determined the extension of the intralabial artery with ultrasonography, including pulsed and color Doppler analysis. We compared the sonograms to the clinical and histopathologic findings. ***Results.*** Sonograms showed clear enlargement of the labial artery in the 3 cases. The course of the constant-diameter artery was either vertical or oblique from the depth of the lip to the surface of the mucosa. This vascular abnormality was confirmed by histopathologic examination. ***Conclusions.*** Ultrasonography and color Doppler imaging may be useful noninvasive tools for the diagnosis and preoperative evaluation, as well as the follow-up, of labial lesions related to a CPLA, thus eliminating the need for diagnostic surgery in typical pulsatile nodules. Ultrasonography may help distinguish a CPLA from other vascular lesions of the lip such as an aneurysm. Atypical cases or chronic ulcerations mimicking a cancer should undergo biopsy so that a malignant process is not missed.
Key words: caliber-persistent artery; labial artery; lip diseases; ulcer; ultrasonography.

Abbreviations
CPLA, caliber-persistent labial artery

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Voth[1] used the term "caliber-persistent artery" in 1962 to describe a primary arterial branch that extended from the base of the gastric wall through the muscular layer and into the submucosa with no reduction of its caliber. These lesions have also been called submucosal arterial malformation, Dieulafoy disease, cirsoid aneurysm, and exulceratio simplex and may be found in the esophagus, duodenum, jejunum, ileum, and colon. These aberrant blood vessels may lead to severe gastrointestinal hemorrhages.[2–6] Since the first description of a "prominent inferior labial artery" by Howell and Freeman[7] in 1973 and the histopathologic criteria of a "caliber-persistent artery" of the lower lip by Miko et al[8] in 1980, a growing number of nodular or ulcerated lesions of the lip related to this abnormal artery have been published. Lovas et al[10] reported a prevalence of 5.3% among 3039 patients referred for clinical oral pathologic consultation. Although no large studies have been performed on the general population, this lesion is now considered a relatively common unilateral vascular

abnormality occurring predominantly on the lateral border of the lower lip.[7,9–12]

Ultrasonography is frequently used for diagnosis, interventional monitoring, and follow-up in many medical fields, including maxillofacial and ear, nose, and throat surgery. Intracranial and extracranial vascular malformations and other vascular anomalies of the head and neck are assessed with ultrasonography and color Doppler imaging.[13–16] Palatal tumors and tongue and floor of the mouth carcinoma are investigated with intraoral ultrasonography before and during surgery, with evaluation of the tumor extension.[17–20]

Currently, practitioners use clinical and histopathologic examination to diagnose a caliber-persistent labial artery (CPLA). We illustrate the use of ultrasonography as a noninvasive diagnostic tool to visualize this abnormal artery. To the best of our knowledge, observations of a CPLA on ultrasonography have not been reported previously.

Material and Methods

We examined the inferior lip of 3 patients with a clinically suspected CPLA. Two female patients had a pulsatile swelling of the lower labial mucosa close to the commissura. The medical history was noncontributory, and there was no history of lip trauma or previous ulceration. The third patient had a chronic nonhealing ulcer of the lower lip vermilion. Carcinoma and a self-inflicted lesion were included in the differential diagnosis.

Before surgery, we localized and determined the extension of the intralabial artery with ultrasonography, including pulsed and color Doppler analysis. We performed the examination with an Acuson system (Siemens Medical Solutions, Mountain View, CA) equipped with a linear probe. The frequency used was 7.5 MHz. With the patient supine, we applied a hydrated polyacrylamide-agar sheet on the patient's lips to prevent distortion of the anatomy from the probe's pressure. After examination of the mucocutaneous junction, we asked the patient to open the mouth to evaluate the vermilion border and labial mucosa of the lower lip. We compared both sides and examined the superficial mucosa, the location, and the course of the labial artery, as well as the aspect of any other blood vessels present in the surrounding soft tissues. The systolic diastolic waveform was analyzed with pulsed Doppler imaging, and a color Doppler record was obtained for each lesion.

For patients with clinical discomfort, we excised the labial lesion after ultrasonographic examination. We also excised the chronic ulceration to rule out a malignant lesion. One symptomless nodule was not removed. We compared the sonograms to the clinical examination and histologic findings. We also performed a follow-up ultrasonographic examination 2 years later.

Case 1

A 52-year-old female patient had a small, soft asymptomatic solitary pulsatile nodule (Figure 1), which had been on the lower lip near the commissura for 2 years. The patient had no clinical discomfort, and an excision was not performed.

Case 2

A 34-year-old female patient had a solitary bluish dome (5 mm in diameter) of the lower lip near the commissura leading to occasional burning discomfort of unknown duration. The nodule was pulsatile, with no underlying induration on palpation.

Case 3

A 54-year-old male patient had a paramedian recurring painful crusty and hemorrhagic ulceration on the lower lip (Figure 2). It was firm, nonpulsatile, and surrounded by moderate cheilitis. There was no visible lesion on the opposite side.

Figure 1. Arrowheads show the contours of a small pulsatile nodule on the left side of the lower lip vermilion (5 mm in diameter) partially extending to the labial mucosa (case 1).

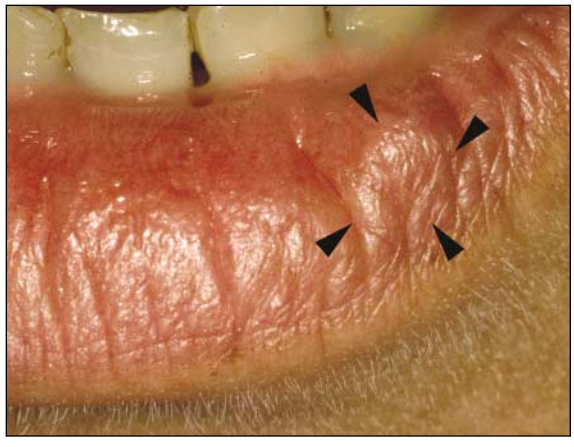


Figure 2. Round ulcer (10 mm in diameter) covered in part by a crusty exudate centered on the swollen lower lip. This recurrent lesion extends to the vermilion border of the lip (case 3).

Results

All sonograms showed the same ultrasonographic pattern (Table 1). The lesions were hypoechoic with no solid or cystic formation of the soft tissues and no signs of necrosis (Figure 3). We observed a regular high peak arterial shift with pulsed Doppler imaging (Figure 4). The waveform of the abnormal artery did not differ from the normal side. Color Doppler analysis clearly showed the enlargement of the labial artery. The course of the constant-diameter artery was either oblique (Figure 5) or vertical from the depth to the surface of the lip. Near the mucosa, the dilated artery showed a V-shape pattern. This pattern was related to a secondary arterial branch in case 1. In case 2, the patient had a nodule where 2 fine blood vessels showed the V-shape arrangement around the dilated artery. One blood vessel was an artery and the other one a vein with no signs of an arteriovenous fistula. The opposite side of the lip was clinically normal, and we observed no architectural changes of the parenchyma; the normal artery was only visible in the depth of the lip. In case 3, the patient had a chronic ulceration, which showed an enlarged artery supplying a large superficial arterial maze with turbulent flow but no signs of an arteriovenous fistula (Figure 6). On the opposite normal side, this patient had a small hypoechoic zone with no arterial signal or abnormal vessel. Ligation and excision of the abnormal vessel were performed without complication, and recovery after surgery was the standard evolution. Histologic examination confirmed the diagnosis of a CPLA.

Two years later, the asymptomatic nodule, which was not excised (case 1), had the same ultrasonographic and clinical aspects. Where the labial lesions were removed, we observed no residual ultrasonographic and clinical anomalies. On the preexistent small hypoechoic zone (case 3), an ulceration had developed. This lesion was contralateral to the initial abnormality. A large hypoechoic zone filled the whole thickness of the lip under the ulceration. We observed a large arteriovenous maze with a turbulent flow (color Doppler and pulsed Doppler imaging).

Table 1. Results of the Initial Sonograms Compared to the Histopathologic Findings

Case	Initial Sonograms	Histopathologic Findings
1: 52-year-old female patient (Figure 1)	No solid or cystic formation in the soft tissues; the extremity of the dilated artery had a constant 1.9-mm diameter and a 9-mm-long vertical course; it composed the bulging of the lip	In agreement with the patient, who had no clinical discomfort, an excision was not performed
2: 34-year-old female patient	Discrete architectural change of the lip with a hypoechoic zone (5 mm in diameter) and no signs of necrosis or nodular mass; visualization of the vertical intralabial course of an enlarged artery with a systolic diastolic waveform (Figure 4)	Intact mucosal surface with discrete submucosal inflammation; presence of dilated artery (0.9 × 1.15 mm) showing intimal thickening with atherosclerotic changes
3: 54-year-old male patient (Figure 2)	Large hypoechoic zone (15 mm in diameter) under the ulceration (Figure 3); visualization of a superficial arterial maze (10 mm in diameter) (Figure 6) supplied by a 15-mm-long dilated artery with an oblique intralabial course (Figure 5); on the opposite side, the hypoechoic zone (5 mm in diameter) had no arterial signal and no abnormal vessel	A crust partially covered the ulceration; presence of an intense submucosal inflammation; a thrombosed branch of a large superficial artery was located above the muscular layer; normal morphologic characteristics of the enlarged artery (0.9 mm in diameter with a maximum of 0.7 × 1.45 mm)

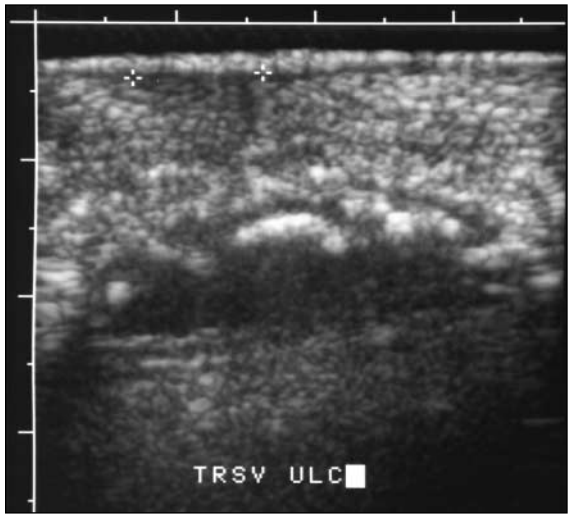


Figure 3. Poorly outlined hypoechoic aspect with no solid or cystic formation of the soft tissues (case 3).

Discussion

Extraoral ultrasonography and a Doppler flow meter are used to evaluate the labial muscle thickness and the activity of a repaired cleft lip, as well as the localization of facial arteries to avoid damaging the arterial pedicle during reconstructive flap surgery.[21–23] To our knowledge, the use of ultrasonography for the diagnosis of a CPLA has not been reported so far.

The vascular anatomy of the labial arteries has been extensively described in human cadaver anatomic studies and in recent histologic studies (Figure 7). Their location, course, and size are subject to great variation.[24,25] For some authors,[22] there is a dual vascular distribution of the upper lip through a superficial and deep septal branch from the superior labial artery, whereas for other authors,[26] there is only a single superior and a single inferior labial artery, which both come close to the free margin of the lip. In another study,[24] the inferior labial artery is the main artery of the lower lip, and it is most frequently found unilaterally rather than bilaterally. The external diameter of a normal inferior labial artery may vary from 0.28 to 1.7 mm and may be different on each side.[8,22,24] These vessels are, in most cases, located on the inner side of the orbicularis oris muscle, but aberrant enlarged arteries traversing the muscle border and continuing "without reduction of caliber in the vicinity of the vermilion border" have been observed in a histologic study.[8] To our knowledge, the intralabial course of a caliber-persistent artery has not yet been described in vivo. We visualized the superficial location of the enlarged artery and its vertical or oblique course in the lip. The echo pattern reflected the structure of the vascular lesion and was in accordance with the histologic findings. Although shrinkage and deformation related to preparation procedures have to be taken into account, our microscopic measurements confirmed the enlarged diameter of the arteries and had histologic features, size, and location similar to those of specimens reported by others.

The 2 clinical patterns of a CPLA observed in our patients were the same as the patterns of a

Figure 4. Systolic diastolic waveform of the enlarged labial artery with a vertical course (case 2).

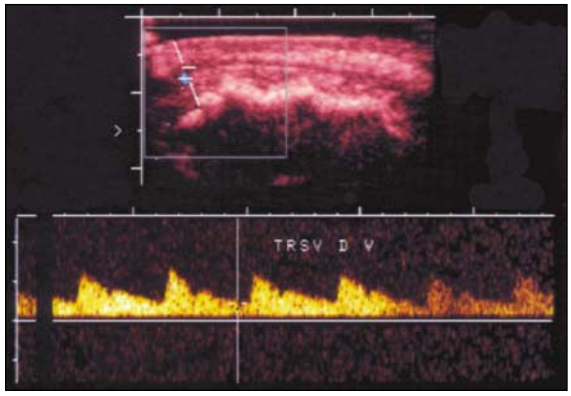


Figure 5. Color Doppler analysis showing a 15-mm-long dilated artery with an oblique intralabial course from the depth of the lip to the surface of the mucosa (case 3).

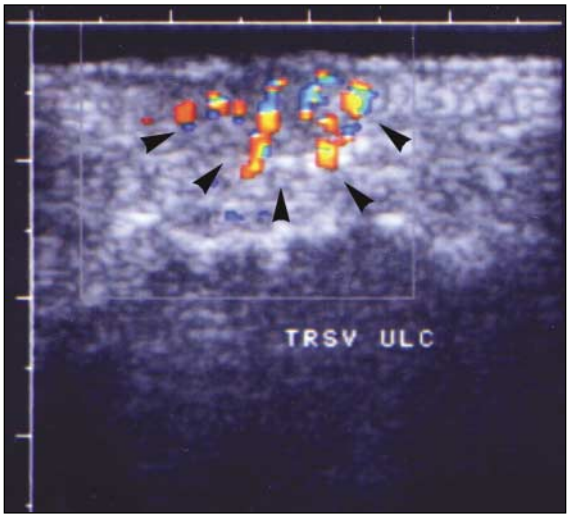
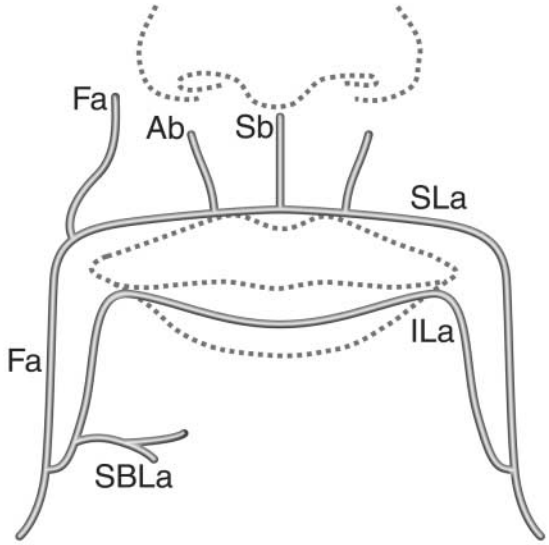


Figure 6. Arrowheads show the superficial arterial maze supplied by the enlarged artery. There are no signs of an arteriovenous fistula on pulsed Doppler imaging (case 3).

CPLA previously described in the literature. The first and common clinical pattern is a unilateral pulsatile submucosal nodule of the vermilion border of the lower lip mucosa.[7,27,28] This pulsatile lesion can usually be clinically distinguished from other nodular lesions of the lip,[11,26–28] and no complementary examination is required. If in doubt or when the patient is anxious, ultrasonography may prevent unnecessary diagnostic surgery. With ultrasonography, we clearly observed the enlarged artery showing a systolic diastolic waveform and confirmed the diagnosis of a CPLA. This tool may also be useful in distinguishing other vascular lesions of the lip such as an aneurysm.[29] We have observed an aneurysm of the inferior lip with ultrasonography (not shown).

Figure 7. Schematic overview of the regional arterial vascularization of the lips. Ab indicates alar branch of the superior labial artery; Fa, facial artery; ILa, inferior labial artery; Sb, septal branch of the superior labial artery; SBLa, sublabial artery; and SLa, superior labial artery. Adapted from Edizer et al,[24] Magden et al,[25] and Schulte et al.[26]



The second pattern is a chronic ulceration related to the CPLA. This lesion, rarely appearing as a paramedian erosion, is usually localized in the Klein zone with no cutaneous involvement and may be responsible for repeated bleeding.[8,9,11,30] The age of the patient and the chronicity of the lesion are important elements because the pulsation of the superficial enlarged artery could be missed when atherosclerotic changes of the vessel occur.[27] Factors such as local trauma of the lip, senile atrophy, tobacco, sun exposure, ischemia of the submucosa covering the enlarged artery, and systolic arterial pressure interfering with the filling of adjacent blood vessels may contribute to the development of this chronic ulceration.[8,21,31,32] The differential diagnosis of a chronic labial ulcer includes several entities, the main being squamous cell carcinoma[8,11,30]; interestingly, the first cases of ulceration related to a CPLA were clinically misdiagnosed as malignant lesions.[8,31] In the presence of a nonhealing labial ulcer of unknown cause, one should think about a CPLA as the initiating factor. Ultrasonography revealing an enlarged artery could incite a careful serial examination of the specimen. Association of a caliber-persistent artery of the stomach with gastric cancer and association of a CPLA with labial squamous cell carcinoma have also been reported.[9,31–33] An excision should therefore be performed when a chronic ulceration of the lip is present.

We primarily used ultrasonographic examination for the diagnosis of a CPLA and suggest that it might be possible to detect different stages of this lesion. We initially observed on the opposite side of the chronic ulcer (case 3) a small hypoechoic zone with no arterial signal and no abnormal vessel. This zone clinically corresponded to a symptom-free area of the lip. An ulceration developed 2 years later at this site with a corresponding large arteriovenous maze but no clear echo pattern of an abnormal artery. We also reexamined the labial asymptomatic nodule 2 years after the initial examination. We observed no

clinical changes, and the ultrasonographic pattern was identical with no increase of the enlarged artery. Ultrasonography may be a useful noninvasive tool for the follow-up examination of either a preexisting well-formed enlarged artery or a possible early stage of a vascular anomaly.

Despite the limited number of investigated patients, ultrasonography and color Doppler imaging show promising preliminary results in labial lesions related to a CPLA and eliminate the need for diagnostic surgery in typical pulsatile nodules.

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