대한영상의학회지

Sonographic Appearance of Steatocystoma: An Analysis of 14 Pathologically Confirmed Lesions

지선낭종의 초음파 소견: 조직학적으로 진단된 14개 병변의 분석

Hyeyoung Yoon, MD (iD), Yusuhn Kang, MD* (iD), Hwiryong Park, MD (iD),
Joong Mo Ahn, MD (iD), Eugene Lee, MD (iD),
Joon Woo Lee, MD (iD), Heung Sik Kang, MD (iD)

Department of Radiology, Seoul National University Bundang Hospital, Seongnam, Korea

*Corresponding author
Yusuhn Kang, MD
Department of Radiology,
Seoul National University
Bundang Hospital,
82 Gumi-ro 173beon-gil,
Bundang-gu, Seongnam 13620,
Korea.

Tel 82-31-787-7619
Fax 82-31-787-4011
E-mail yskang0114@gmail.com

ORCID iDs
Hyeyoung Yoon (iD)
https://orcid.org/0000-0002-9913-6885
Yusuhn Kang (iD)
https://orcid.org/0000-0003-1838-2564
Hwiryong Park (iD)
https://orcid.org/0000-0001-9830-9097
Joong Mo Ahn (iD)
https://orcid.org/0000-0002-1157-0020
Eugene Lee (iD)
https://orcid.org/0000-0003-4205-2362
Joon Woo Lee (iD)
https://orcid.org/0000-0002-7106-5229
Heung Sik Kang (iD)
https://orcid.org/0000-0002-7024-388X

Purpose To evaluate the ultrasonographic characteristics of steatocystomas focusing on the features that aid in differentiating them from epidermal inclusion cysts and lipomas.

Materials and Methods The ultrasonographic findings of 14 histologically proven steatocystomas in 10 patients were retrospectively reviewed. The following features were assessed: the layer of involvement, shape, margin, echogenicity, posterior acoustic features, and the presence of a visible wall or intralesional striations. The findings were compared with those of subcutaneous lipomas and epidermal inclusion cysts to identify those findings that aid in the differential diagnosis of steatocystomas.

Results The majority of steatocystomas appeared as a subcutaneous mass (n = 6, 42.9%) or a mass involving both the dermal and subcutaneous layers (n = 6, 42.9%). Steatocystomas exhibited a well-defined smooth margin (n = 12, 85.7%) and homogeneous echogenicity (n = 9, 64.3%), and showed no specific posterior acoustic features (n = 9, 64.3%). The most important features that differentiated steatocystomas from epidermal inclusion cysts were a homogeneous internal echotexture (p = 0.009) and absent or less prominent posterior acoustic enhancement (p < 0.001). The features that distinguished steatocystomas from lipomas were the margin (p < 0.001), echogenicity (p = 0.034), internal echotexture (p = 0.004), and the absence of intralesional striations (p < 0.001).

Conclusion Steatocystomas appeared as well-defined homogeneous masses with mild or absent posterior acoustic enhancement.

Index terms Steatocystoma Multiplex; Soft Tissue Neoplasm; Ultrasonography

INTRODUCTION

Steatocystoma is a lesion that results from a hamartomatous malformation of the pilosebaceous duct, leading to an epithelium-lined cystic lesion containing sebaceous lobules (1). Steatocystomas may occur anywhere in the body, but are more commonly reported where pilosebaceous units are well-developed, including the trunk, neck, axillae, groin, scalp, and proximal extremities. The incidence of steatocystoma is unknown, but is considered a rare lesion that equally affects both sexes (2). They are known to appear during adolescence suggesting a possible hormonal trigger (3, 4).

Steatocystomas can be classified into steatocystoma multiplex and steatocystoma simplex, according to the multiplicity. Steatocystoma multiplex is known to be an autosomal dominant disorder, but sporadic cases have been reported. Recent studies have also revealed that steatocystoma multiplex is associated with mutations in the keratin 17 (K17) gene (5, 6).

Steatocystomas usually manifest as solitary or multiple palpable nodules that are skin-colored and asymptomatic. The diagnosis of steatocystoma multiplex may be performed clinically when intradermal lesions occur in typical locations with multiplicity, when there is a family history (7). However, steatocystoma simplex and sporadic cases of steatocystoma multiplex that manifest as subcutaneous palpable lesions may pose a diagnostic challenge. Clinically, the presentation of these lesions may be similar to those of other more common benign subcutaneous masses such as epidermal inclusion cysts (EICs), lipomas, or lipoma-variants.

The ultrasonographic findings of steatocystomas have been reported in a limited number of studies (7-11), three of which reported mammographic and ultrasonographic findings of steatocystoma multiplex occurring in the breast. Park et al. (10) reported the ultrasound findings of steatocystoma multiplex in four patients; sonographic findings revealed multiple well-circumscribed homogeneous lesions that either confined to the dermal layer or involved the subcutaneous fat.

Based on our clinical experience, we noted that steatocystomas have been erroneously assessed as other subcutaneous masses both clinically and sonographically. Thus, the purpose of this study was to evaluate the ultrasonographic characteristics of steatocystomas, focusing on the features that aid in the differential diagnosis of steatocystomas from EICs and lipomas.

MATERIALS AND METHODS

This retrospective study was approved by our Institutional Review Board, and informed consent was waived (IRB No. B-1904/532-108).

PATIENT SELECTION

A search of the electronic medical records revealed 17 patients that visited our hospital between May 2005 and June 2017, who 1) had been clinically diagnosed with either steatocystoma multiplex or simplex, and 2) had undergone ultrasound examination. Among these patients, we selected those who had undergone surgical excision of the lesion and were

histologically proven to have steatocystoma. A total of 14 pathologically proven steatocystomas from 10 patients were included in our study. Among the 10 patients, six were diagnosed with steatocystoma multiplex and four with steatocystoma simplex. There were five female patients (mean age, 39.2 years, range 26–51 years) and five male patients (mean age, 53.8 years, range 46–64 years), with an overall mean age of 46.5 years (range, 26–64 years).

CONTROL GROUP SELECTION

On retrospective review of the initial radiologic reports of ultrasound examinations in our institution, we found that steatocystomas had been erroneously assessed as either EICs or lipomas in a majority of cases. In order to identify ultrasonographic findings that aid the differential diagnosis of steatocystomas from EICs and lipomas, we selected cases of EICs and lipomas as the control group. We searched our electronic medical record between May 2005 and June 2017 to identify patients who had undergone ultrasound examination and was confirmed with either lipoma or EIC on subsequent histologic examination. There were 148 histologically confirmed lipomas and 104 histologically confirmed EICs. Only lesions with size equal to or less than 20mm were selected, considering the small size of the included steatocystomas. Among the lesions fulfilling the size criterion (44 lipomas and 69 EICs), 14 lipomas and 14 EICs were randomly selected based on a random number generated with Microsoft Excel. The lipoma group included 6 females and 8 males with a mean age of 51.5 years (range, 29–70 years), whereas the EIC group included 7 females and 7 males with a mean age of 44.7 years (range, 23–80 years).

SONOGRAPHIC EXAMINATIONS AND IMAGE ANALYSIS

Ultrasonographic examinations were performed by 23 radiologists with 1–9 years of experience in musculoskeletal ultrasonography. Imaging was performed with either an iU22 scanner ($n = 11$) or an Epic7 scanner ($n = 3$) using a 5–12 MHz, 5–17 MHz, or 5–18 MHz linear array transducer (iU22 and EPIQ 7, Philips Healthcare, Best, the Netherlands).

Three readers who did not perform the ultrasonographic examinations retrospectively reviewed the acquired images. First, two readers (Y.K. and H.Y. with 1 and 9-year experience in ultrasonography) independently reviewed the images, blinded to the pathologic report. Discrepancies between the two readers were resolved by a third reader (H.P. with 5-year experience in ultrasonography).

The following features were evaluated; 1) location of lesion (intradermal, subcutaneous layer, involving both dermal and subcutaneous layers), 2) shape of lesion (round or oval, spindle-shaped, irregular), 3) margin of lesion (ill-defined, well-defined smooth, well-defined irregular), 4) echogenicity of lesion, 5) internal echotexture, 6) presence of intralesional striations, and 7) posterior acoustic features. A lesion with well-defined smooth margin shows an abrupt transition between lesion and surrounding tissue, with an even, gradually curving interface. A well-defined irregular margin refers to lesions with an abrupt transition between lesion and surrounding tissue but shows lobulation, angulation or speculation, Lesions without clear demarcation between the mass and its surrounding tissue were categorized as having an "ill-defined margin." The echogenicity of the lesions was analyzed with reference to the echogenicity of the dermal and subcutaneous layers, and was classified into 4 levels: hy-

poechoic to fat, isoechoic to fat, hyperechoic to fat but hypoechoic to the dermis, and isoechoic or hyperechoic to the dermis. The internal echotexture of the lesion was assessed as homogeneous or heterogeneous, and the presence of intralesional hyperechoic striations was also recorded. The posterior acoustic features of the lesions were divided into four categories: no posterior acoustic feature (no posterior acoustic enhancement or shadowing), mild posterior acoustic enhancement (a slight increase in echogenicity of the area posterior to the mass, compared to adjacent tissue at the same depth), strong posterior acoustic enhancement (marked increase in echogenicity of the area posterior to the mass, compared to adjacent tissue at the same depth), and posterior acoustic shadowing (the area posterior to the mass appears hypoechoic compared with the adjacent tissue).

STATISTICAL ANALYSIS

Demographic data and imaging findings were summarized using descriptive statistics, including means, ranges, and frequencies, as appropriate. Fisher's exact test was performed to assess the ultrasonographic findings that aid differentiation between steatocystomas and EICs and steatocystomas and lipomas. To evaluate the interobserver agreement between the two radiologists that assessed the sonographic features of the soft tissue masses, Cohen's kappa statistics were used and the weighted kappa value was obtained to determine the echogenicity, which was considered an ordinal variable. Kappa values were evaluated according to the following classification: 0-0.20, poor agreement; 0.21–0.40, fair agreement; 0.41–0.60, moderate agreement; 0.61–0.80, good agreement; 0.81–1.0, excellent agreement. All statistical analyses were performed with SPSS (version 24.0, IBM Corp., Armonk, NY, USA) and MedCalc (version 18.10.2, Ostend, Belgium). A p value of < 0.05 was considered statistically significant.

RESULTS

A summary of the clinical findings is shown in Table 1. Lesions were located in the trunk ($n = 6$), upper extremity ($n = 4$), axilla ($n = 2$), and the head and neck region ($n = 2$). Using ultrasound examination, the first diagnostic consideration was lipoma or a lipoma-variant in six lesions, EIC in four lesions, neurofibromas in two lesions, and other benign masses in the remaining two lesions.

SONOGRAPHIC FINDINGS OF THE STEATOCYSTOMAS

The mean size of the steatocystoma was 7.9 mm; range: 4 mm to 14 mm. Six lesions (42.9%) were located within the subcutaneous layer (Fig. 1), two lesions (14.3%) were limited to the dermal layer (Fig. 2A), and six lesions (42.9%) involved both dermal and subcutaneous layers (Fig. 3). Lesions were round or oval ($n = 12$), spindle-shaped ($n = 2$), and irregular-shaped ($n = 1$). The majority of lesions ($n = 12$, 85.7%) had a well-defined smooth margin, whereas one lesion had a well-defined irregular margin, and another exhibited an ill-defined margin. Three lesions (21.4%) were hypoechoic to subcutaneous fat (Fig. 4), six lesions (42.9%) were isoechoic to subcutaneous fat (Fig. 3), three lesions (21.4%) were hypoechoic to subcutaneous fat (Fig. 4), and five (35.7%) were hyperechoic to subcutaneous fat, but hy-

Table 1. Demographic Data and Clinical Information of Patients with Steatocystomas

Patient	Age/Sex	Location	Size (cm)	US Diagnosis	Final Clinical Diagnosis
#1	54/M	Left arm	1.0	Lipoma-variant	SM
#2	64/M	Eyebrow	1.2	Complicated EIC	SS
#3	42/F	Left forearm	0.6	Dermatofibroma	SS
#4	58/M	Right arm	1.3	Lipoma	SM
		Left arm	0.7	Lipoma	
#5	47/M	Back#1	0.6	EIC	SM
		Back #2	0.4	EIC	
		Back #3	0.5	EIC	
#6	46/M	Right axilla	1.4	Lipoma with cystic degeneration	SS
#7	48/F	Neck	0.7	Lipoma-variant	SM
#8	51/F	Abdominal wall	0.8	Lipoma	SM
#9	29/F	Left axilla	0.7	Benign lesion	SS
#10	26/F	Abdominal wall	0.4	Neurofibromatosis	SM
		Abdominal wall	0.6	Neurofibromatosis	

EIC = epidermal inclusion cyst, SM = steatocystoma multiplex, SS = steatocystoma simplex, US = ultrasound
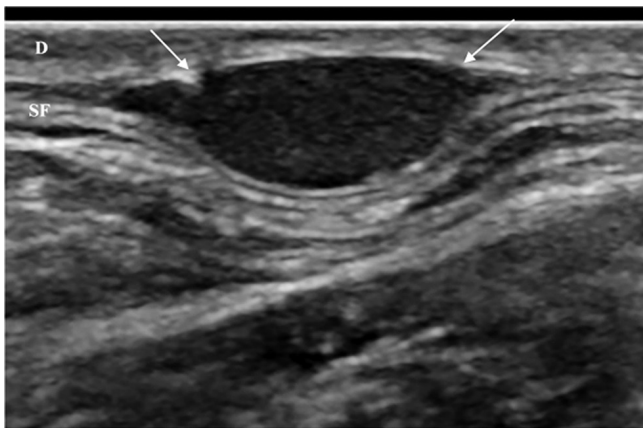
Fig. 1. A steatocystoma in a 48-year-old female who had a palpable mass in the right anterior neck.
The sonogram shows a well-defined spindle-shaped hypoechoic mass in the SF. The hyperechoic interface between the D and SF is smoothly marginated (arrows). The lesion was initially assessed as a lipoma-variant.
D = dermis, SF = subcutaneous fat

poechoic to the dermal layer (Fig. 2B). The internal echogenicity was homogeneous in nine lesions (64.3%), whereas a heterogeneous echotexture was noted in five lesions (35.7%). Only one lesion exhibited intralesional striations. Mild posterior acoustic enhancement was noted in five lesions (35.7%) (Fig. 2A), whereas no specific posterior acoustic feature was noted in the other nine lesions (64.3%).

COMPARISON OF THE SONOGRAPHIC FEATURES OF THE STEATOCYSTOMAS WITH THOSE OF EICS AND LIPOMAS

The ultrasonographic features of steatocystomas in comparison with EICs and lipomas are summarized in Table 2. Steatocystomas mostly showed no specific posterior acoustic feature or mild posterior acoustic enhancement, whereas EICs mostly showed strong posterior acoustic enhancement (Fig. 5), and the difference was statistically significant ($p < 0.001$). In addition, steatocystomas mostly showed a homogeneous echotexture, while epidermoid in-

Fig. 2. Steatocystomas in a 47-year-old male who had multiple masses at the back.
A. The sonogram shows a well-defined oval hypoechoic mass in the D with mild posterior acoustic enhancement (arrows).
B. Sonogram of another lesion shows a well-defined oval mass with echogenicity higher than the SF but lower than the D, with mild posterior acoustic enhancement. Initially, both lesions were erroneously assessed as epidermal inclusion cysts.
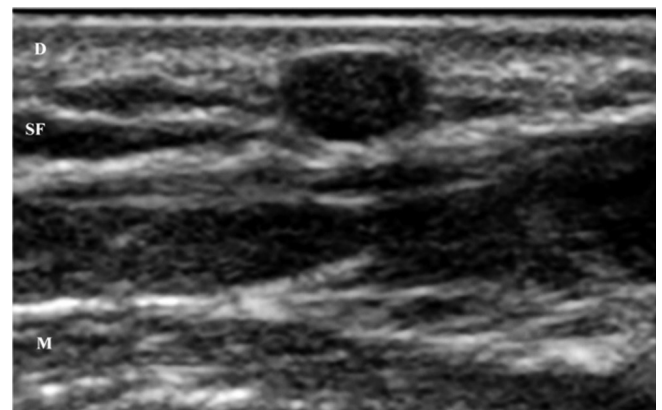D = dermis, M = muscles, SF = subcutaneous fat
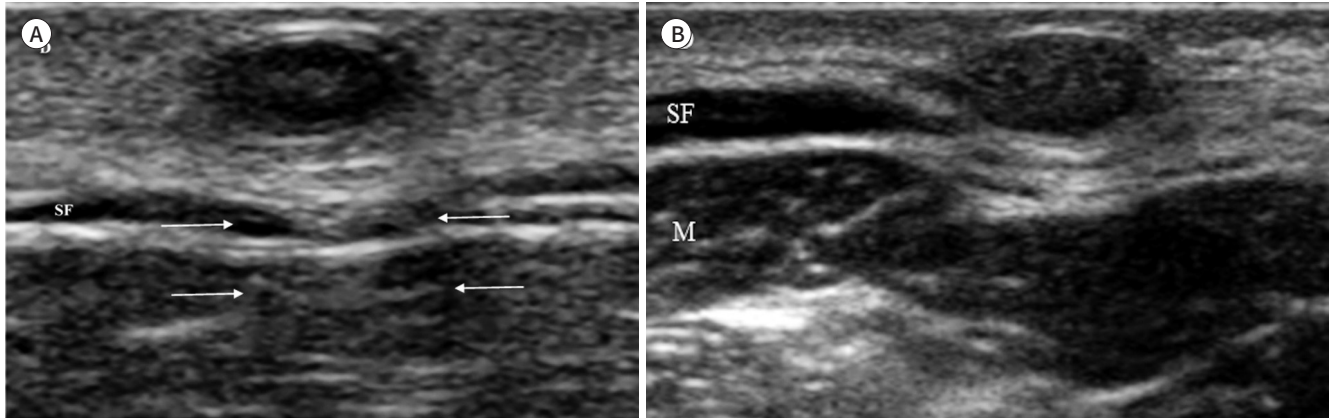
Fig. 3. A steatocystoma in a 58-year-old male who had multiple palpable masses in the left forearm.
The sonogram shows a well-defined oval mass involving both the D and SF layers. The lesion appears isoechoic to the SF layer. Initially, the lesion was erroneously assessed as a lipoma.
D = dermis, M = muscles, SF = subcutaneous fat

Fig. 4. A steatocystoma in a 42-year-old female who presented with a palpable mass in the left forearm, which was initially assessed as a dermatofibroma.
The sonogram shows a well-defined mass in the SF with echogenicity hypoechoic to the adjacent SF. The lesion shows no posterior acoustic enhancement.
D = dermis, M = muscles, SF = subcutaneous fat
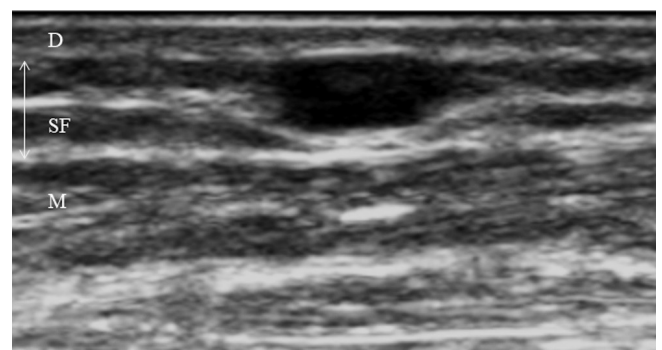
clusion cysts mostly showed a heterogeneous echotexture ($p < 0.009$). Other features, including the location, shape, margin, and echogenicity of lesions and intralesional striation did not show statistically significant differences between the two groups.

A larger proportion of the lipomas showed the following features, compared to steatocystomas, with statistically significant differences: 1) ill-defined margin ($p < 0.001$), 2) hyperechogenecity to subcutaneous fat ($p = 0.0034$), 3) heterogeneous internal echotexture ($p = 0.004$),

Table 2. Ultrasound Features of Steatocystomas Compared to EICs and Lipomas

	Steatocystoma (n = 14)	EIC (n = 14)	p-Value	Lipoma (n = 14)	p-Value
Location			0.871		0.059
Intradermal	2	2		0	
Subcutaneous	6	4		12	
Involving both layers	6	8		2	
Shape			0.730		0.078
Round or oval	11	13		5	
Spindle-shaped	2	0		7	
Irregular	1	1		2	
Margin			0.596		< 0.001
Ill-defined	1	0		11	
Well-defined smooth	11	13		2	
Well-defined irregular	2	1		1	
Echogenicity of the lesion			0.247		0.034
Hypoechoic to fat	3	7		0	
Isoechoic to fat	6	3		3	
Hyperechoic to fat, Hypoechoic to dermis	5	3		7	
Isoechoic or hyperechoic to dermis	0	1		4	
Internal echotexture			0.009		0.004
Homogeneous	9	2		1	
Heterogeneous	5	12		13	
Intralesional striations			1.000		< 0.001
Absent	13	14		3	
Present	1	0		11	
Posterior acoustic feature			< 0.001		0.678
Absent	9	1		11	
Mild posterior acoustic enhancement	5	3		3	
Strong posterior acoustic enhancement	0	10		0	
Posterior acoustic shadowing	0	0		0	

Data are number of patients, unless indicated otherwise. p values are for comparison between groups with Fischer exact test.
EIC = epidermal inclusion cyst

and 4) intralesional striations ($p < 0.001$) (Fig. 6).

INTEROBSERVER AGREEMENT BETWEEN THE TWO READERS IN EVALUATING THE SONOGRAPHIC FEATURES OF THE SOFT TISSUE MASSES

The interobserver agreement between the two readers was fair to moderate. The kappa co-efficient for each variable was calculated as follows: 1) location ($\kappa = 0.52$), 2) shape ($\kappa = 0.30$), 3) margin ($\kappa = 0.55$), 4) echogenicity of the lesion (weighted $\kappa = 0.51$), 5) internal echotexture ($\kappa = 0.46$), 6) intralesional striations ($\kappa = 0.50$), and 7) posterior acoustic feature ($\kappa = 0.57$).

Fig. 5. An epidermal inclusion cyst in a 38-year-old male who had a palpable mass in the left lower back.
The sonogram shows a well-defined heterogeneously hypoechoic mass involving both the D and SF layers. This lesion shows strong posterior acoustic enhancement (arrows) and internal echogenic reflections that may correspond to the lamellated keratinous material.
D = dermis, SF = subcutaneous fat
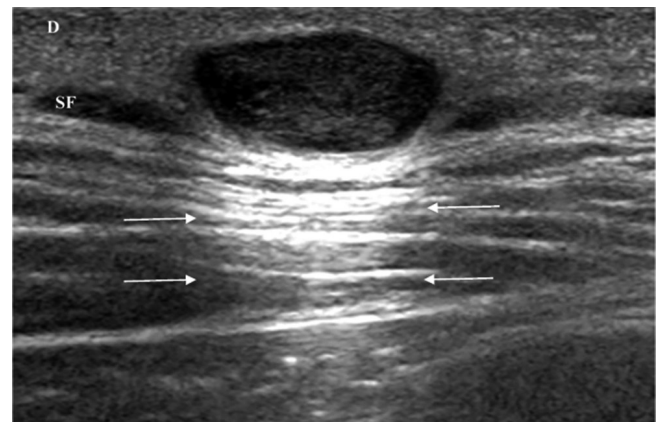
Fig. 6. A lipoma in a 57-year-old male who had a growing mass in the left upper arm.
The sonogram shows an elongated mass (L) with isoechogenecity to the adjacent SF. This lesion shows intralesional striation (arrowhead), which is a key feature that differentiates lipomas from steatocystomas.
M = muscles, SF = subcutaneous fat
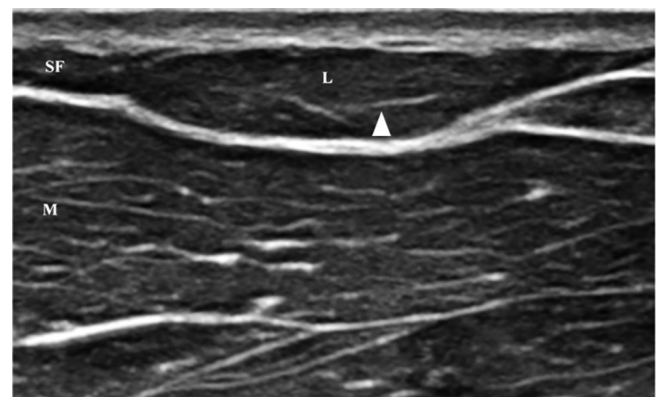
DISCUSSION

In our study, steatocystomas showed a well-defined smooth margin, homogeneous internal echotexture, and no specific posterior acoustic feature or mild posterior acoustic enhancement. The majority of cases in our study were initially diagnosed as lipomas, EICs, and neurofibromas. This aligns with previous reports that have shown that steatocystomas may be clinically confused with EICs, lipomas, xanthomatosis, neurofibromatosis, and syringomas (12, 13). Many benign subcutaneous masses are considered as nonspecific solid masses and require biopsy or excision for definitive diagnosis (14). The results of our study may aid in the differential diagnosis of steatocystomas form other benign subcutaneous masses based on ultrasonographic findings.

Ultrasonography can play an important role because subcutaneous lesions can be difficult to differentiate based on clinical findings. Park et al. (10) reported the ultrasound findings of 18 steatocystomas in four patients: sonographic findings revealed multiple well-circumscribed oval hypoechoic lesions with posterior enhancement. They reported that all 18 lesions appeared homogeneous internally, which is consistent with the results of our study. In their study, the lesions were either confined to the dermal layer (44.4%) or showed involvement of both dermal and subcutaneous fat layers (55.6%). However, in our study, only 2 lesions were confined to the dermal layer and the others were appeared as a subcutaneous mass or a mass involving both dermal and subcutaneous layers. This discrepancy may have resulted from a

selection bias in the inclusion of patients for our study, as typical multiple intradermal steatocystomas were not referred for ultrasound examination. A recent report by Zussino et al. (11) showed that steatocystomas may appear as hypoechoic nodules with well - defined hyperechoic borders with absence of Color Doppler signal. Furthermore, posterior acoustic enhancement could be seen when the internal content was liquid. This is consistent with the results of our study: 92.9% of the steatocystomas showed well-defined borders and 35% showed mild posterior acoustic enhancement. The well-defined borders of steatocystomas are attributable to the histologic feature of steatocystomas: the cyst walls are composed of three to five layers of squamous epithelium and are lined with flattened sebaceous glands (15, 16).

In our study, steatocystomas mostly showed a homogeneous echotexture and absent or mild posterior acoustic enhancement, whereas EICs showed a heterogeneous internal echotexture with strong posterior acoustic enhancement. The ultrasonographic features of EICs found in the present study are consistent with previously reported findings (17-20). Huang et al. (19) reported a pseudotestis appearance of EICs, with intralesional bright echogenic reflectors and filiform anechoic areas. This coincides with the heterogeneous internal echogenicity noted in EICs in our study. EICs are known to show a strong posterior acoustic enhancement (17, 18). This was found to be an important feature that differentiate EICs from steatocystomas, in our study. The internal content of the steatocystomas have been reported to be yellowish-dirty fluid that has acellular, granular debris, rare anucleated squamous cells and rare cholesterol crystals on histologic examination (21). The heterogeneity of the internal content of steatocystomas may have possibly resulted in a less prominent posterior acoustic enhancement compared to EICs. On encountering a subcutaneous mass with homogeneous echotexture and absent or mild posterior acoustic enhancements on ultrasound, one should consider the possibility of a steatocystoma rather than an EIC.

Steatocystomas differed from lipomas in terms of location, internal echotexture, and the presence of intralesional striations. Steatocystomas were present in variable locations and exhibited a homogeneous internal echotexture, but lipomas were almost invariably located in the subcutaneous fat layer and showed a heterogeneous internal echotexture. The typical ultrasound findings of lipoma are well-circumscribed mass with variable echogenicity compared with the surrounding soft tissue (14). Although lipomas usually have a well-defined appearance with an identifiable thin capsule, some may have an ill-defined margin that blends with the adjacent subcutaneous fat (22-24). In our study, lipomas were more frequently assessed to have ill-defined margins compared to steatocystomas. Our study results coincide with the study by Inampudi et al. (23), in which the majority of lipomas had ill-defined borders; 52–60% of the lipomas included in their studies were assessed to have ill-defined borders. Another characteristic feature of lipomas is that curved echogenic lines can be noted within the mass (14). This was one of the key features that differentiated lipomas from steatocystomas in our study.

Several limitations of our study are noteworthy. First, due to the retrospective design of the study, we could not control biases resulting from the use of different ultrasound machines, probes and the differences in the level of experience of the examiners. Second, the study sample was small, and further studies with a larger number of subjects are required. Third, there may be a selection bias in our patient group, as only a number of patients with steatocystoma

in the database underwent ultrasound; those with typical clinical presentations were diagnosed clinically and were not subjected to sonographic evaluation. Fourthly, due to the retrospective nature of the study, advanced ultrasound techniques such as elastography could not be applied. Lastly, the interobserver agreement of the sonographic features were limited in our study. The low agreement could have possibility resulted from the diverse ultrasonographic examination settings, and further analysis with controlled ultrasound examination settings may be necessary.

In conclusion, steatocystomas were noted as well-defined homogeneous masses with mild or absent posterior acoustic enhancement, and were located in the subcutaneous layer or involved both dermal and subcutaneous layers. These findings may aid the differential diagnosis of steatocystomas from other subcutaneous soft tissue masses on ultrasound.

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지선낭종의 초음파 소견: 조직학적으로 진단된 14개 병변의 분석

윤혜영 · 강유선* · 박휘롱 · 안중모 · 이영준 · 이준우 · 강흥식

목적 지선낭종의 초음파 소견을 분석하고, 표피낭종 및 지방종과의 감별진단에 도움이 되는 소견을 알아보고자 하였다.

대상과 방법 10명의 환자에서 14개의 조직학적으로 확진된 지선낭종의 초음파 소견을 후향적으로 검토하였다. 병변의 위치, 모양, 경계, 에코 발생 정도, 후방 음향 특징 및 테두리 벽 또는 병변 내 줄무늬의 존재 여부를 평가하였다. 지선낭종의 초음파 소견을 분석하고, 표피낭종 및 지방종과의 감별진단에 도움이 되는 소견을 알아보고자 하였다.

결과 지선낭종의 대부분은 피하 종괴($n = 6$, 42.9%) 또는 피부층과 피하층을 함께 침범한 종괴로($n = 6$, 42.9%) 나타났다. 병변은 대부분 경계가 잘 지어지고($n = 12$, 85.7%) 균질한 에코 발생을 보였으며($n = 9$, 64.3%), 후방 음향 특징을 나타내지 않았다($n = 9$, 64.3%). 표피낭종을 지선낭종과 구별하는 가장 중요한 소견은 균질한 내부 에코 발생과($p = 0.009$), 현저하지 않은 후방 음향 향상이다($p < 0.001$). 지선낭종과 지방종의 구별되는 특징은 경계($p < 0.001$), 에코 발생성($p = 0.034$), 내부 에코 성상($p = 0.004$) 및 병변 내 줄무늬의 부재였다($p < 0.001$).

결론 지선낭종은 초음파 검사상 경계가 좋고 균질하며, 후방 음향 향상이 경도로 있거나 없는 종괴로 관찰되었다.

분당서울대학교병원 영상의학과