Annals of Diagnostic Pathology 41 (2019) 62–68

Contents lists available at ScienceDirect

Annals of Diagnostic Pathology

journal homepage: www.elsevier.com/locate/anndiagpath

Original Contribution

Benign mesenchymal tumors of the external ear: A series of 14 cases

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ARTICLE INFO

Keywords:
Ear
External
Mesenchymal
Benign
Tumors
Polyps
Nodules

ABSTRACT

Primary soft tissue tumors of the head and neck region are relatively uncommon. Most are not distinctive to this region. Benign mesenchymal tumors of the external ear are rare. Some are common tumors but unusual in this location. All of the reported cases were single case reports or small series. Our aim was to study the prevalence and spectrum of different types of benign mesenchymal tumors that involve the external ear in our institution, to find out whether any lesion is distinctive to this site, their potential clinical associations and to highlight their potential diagnostic challenges. We performed a retrospective review study over 13 years. We retrieved 14 cases of external ear tumors. They included two cases of leiomyomas, two hemangiomas, three neurofibromas, two xanthogranulomas, three osteomas, a lipoma and a sclerotic fibroma. The age range was between 8 and 61 years with an average age of 34.2 years. The male to female ratio was 1.3 to 1. The average size was 8 mm. They were miscellaneous uncommon lesions and most were not unique to the external ear. Meatal osteomas and auricular angioleiomyomas are not infrequent with some predilection to the ear. With the exception of neurofibromatosis type-1, they were solitary nonsyndromic lesions. Multiplicity can be a hint to a syndrome. Clinically, benign external ear mesenchymal tumors can be confused with neoplastic and nonneoplastic lesions. Histopathologic examination is needed for proper classification. Benign soft tissue tumors of the external ear are generally easy histologic diagnosis. Immunohistochemistry is needed to confirm the diagnosis in certain tumors showing overlapping features.

1. Introduction

Soft tissue tumors of the head and neck area are relatively uncommon and mostly malignant [1]. Most mesenchymal neoplasms are not distinctive to this region [1]. Benign mesenchymal tumors are rare, not unique and not primarily localized to the external ear [2,3]. Some are common tumors but unusual in this location [4]. Most of the reported cases were single reports or small series. Pathologists showed a relatively little interest to benign mesenchymal tumors of the external ear as a group compared to tumors in other head and neck sites. Some may present clinical and pathologic diagnostic challenges which necessitate understanding of ear anatomic associations and pathology [4,5]. We performed a retrospective review study to investigate the different types of benign mesenchymal neoplasms of the outer ear in our institution, to investigate whether any is distinctive to this site, their potential clinical associations and to study their potential diagnostic challenges.

2. Materials and methods

We searched for mesenchymal tumors, stromal nodules, soft tissue neoplasms, dermal papules, polyps and lesions that involved the external ear, as well as bony and cartilaginous tumors. We have used a computer-based search to retrieve all of the cases of mesenchymal tumors that primarily involved the external ear in our institution over the past 13 years from 2005 to 2019. We have reviewed the microscopic description, final diagnosis and diagnosis comment of all the pathology reports. We selected cases with an established diagnosis of mesenchymal neoplasm. We have excluded keloids, scarred nodules, pyogenic granuloma, granulation tissue, fibroepithelial polyps, skin tags, inflammatory otic polyps, melanocytic nevi, congenital anomalies and accessories. Periauricular lesions and lymph nodes were excluded. Cases with a secondary extension from the middle ear or through tympanic membrane were also excluded as well as malignant neoplasms. We retrieved the archived hematoxylin and eosin (H&E) stained slides of the selected cases. The slides were reviewed to confirm the diagnosis and to classify the tumors. We have collected the relevant

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https://doi.org/10.1016/j.anndiagpath.2019.05.011

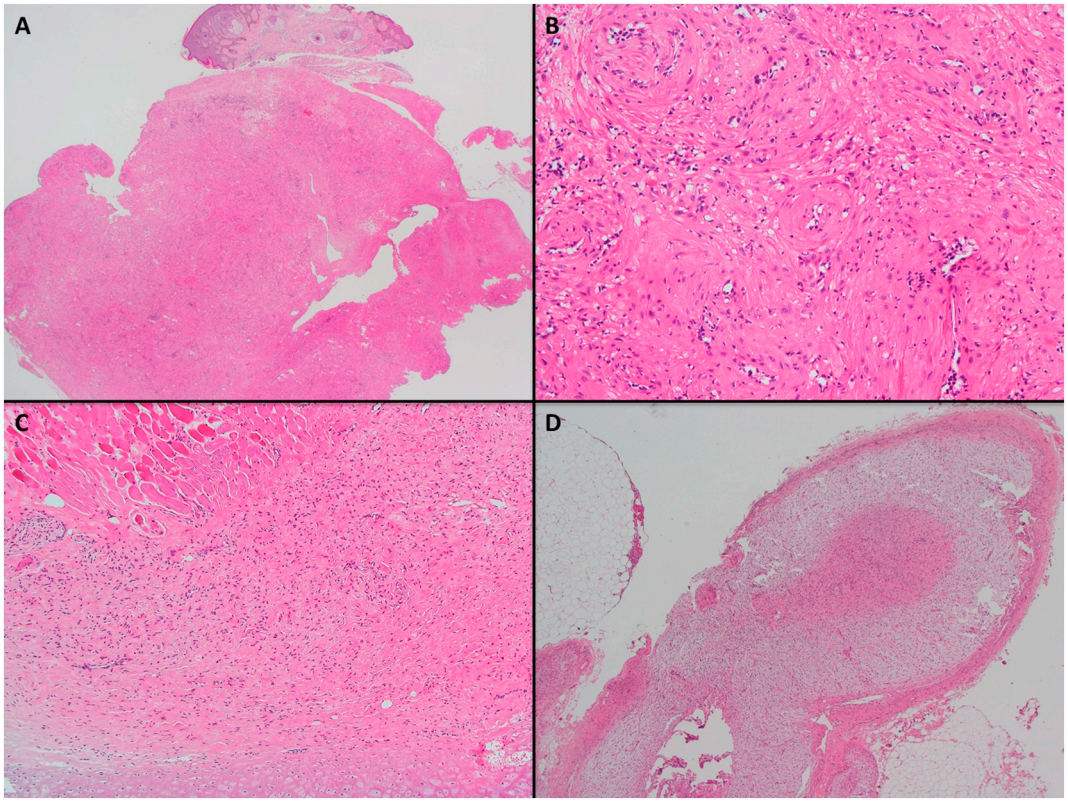

Fig. 1. A) Angioleiomyoma shows a well-defined round solid nodule of the dermis (Hematoxylin and eosin ×20). B) The nodule is composed of interlacing fascicles of proliferating spindle to plump oval cells intimately associated with muscular blood vessels (H&E ×200). C) Deep neurofibroma shows a deep irregular nodule composed of wavy spindle cells involving the skeletal muscle bundles and cartilage (H&E ×400). D) Plexiform neurofibroma composed of an enlarged nerve expanded by myxoid spindle cell proliferation (H&E ×40).

clinical findings for each patient and any available follow up data. The gross, pathologic and pertinent immunohistochemical findings for each case were gathered. We also attempted to find if any other lesions, clinical conditions or syndromes are associated with the external ear mesenchymal tumors.

3. Results

We found 14 cases (4.8%) of benign mesenchymal tumors out of 289 external ear lesions that primarily involved the external ear. The age range was between 8 and 61 years with an average age of 34.2 years. The male to female ratio was 1.3:1. Eleven cases involved the right ear and three the left ear. Six lesions were from the pinna, six from the canal and two from the lobe. Eleven cases were asymptomatic nodules that were present for months to years and the patients sought removal for either cosmetic reasons or due to a recent increase in size. The size ranged from 5 mm to 13 mm with an average size 8 mm. Two cases of neurofibroma were multiple lesions while the other 12 cases were single lesions (Table 1). Two cases of osteoma were associated with psoriasis, one with psoriatic arthritis. Both patients received biotherapy. A third case of osteoma was associated with acanthosis nigricans. A child with plexiform neurofibromas and multiple dermal and deep neurofibromas was found to have café au lait spots and clinical and radiologic features of Recklinghausen disease with a family history of neurofibromatosis from the mother side. The remaining 10 cases were isolated nodules without relevant associated lesions or diseases. The tumors did not recur after excision. The cases included smooth muscle, peripheral nerve sheath, histiocytic, fibroblastic/myofibroblastic, vascular, adipocytic and bony tumors. The smooth muscle tumors included angioleiomyoma and deep leiomyoma, the neurogenic tumors included dermal neurofibromas, deep neurofibromas and plexiform neurofibromas, the histiocytic and fibroblastic tumors included juvenile xanthogranulomas and sclerotic fibroma, and the vascular tumors included capillary hemangioma and arteriovenous hemangioma. Lipoma represented the adipocytic tumors and osteomas the bony tumors. With the exception of osteomas, most were clinically confused with other benign lesions (Table 1). The diagnosis was morphologically easy in most of the cases. A limited panel of immunohistochemistry markers was used in selected cases to confirm the diagnosis (Table 1). The juvenile xanthogranulomas were histologically confused with benign fibrous histiocytoma and Langerhans cell histiocytosis. The cells were positive for CD68, but negative for smooth muscle actin (SMA), factor XIIIa (FXIIIa), S-100, CD1a and CD207 (langerin). They lacked hemosiderin, storiform growth pattern, grenz zone and epidermal changes. The deep leiomyoma showed degenerative and inflammatory changes, and morphologically mimicked deep fibrous histiocytoma and schwannoma. The cells were strongly and diffusely positive for SMA. They were negative for S-100 and CD34. CD45, CD3 and CD68 showed inflammatory lymphohistiocytic background. The superficial dermal neurofibromas were confused with dermatofibromas. The overlying epidermis was unremarkable. The cells were positive for S-100 protein and negative for FXIIIa and SMA.

4. Discussion

The external ear is anatomically divided into the cartilaginous auricle or pinna, the fatty lobe or lobule and the canal or meatus with outer cartilaginous and inner bony parts. Histologically, the different p-

Table 1
Clinical and pathologic features of the external ear nodules.

SN	Age	Sex	Clinical presentation/Topography/Impression	Gross findings and size (mm)	Pathologic findings/Immunohistochemistry	Diagnosis	Associated lesion/Follow up
1	42	M	Painless nodule × 2.5 mths/left earlobe/skin tag, cystic lesion, keloid.	Yellow to pink firm round nodule (9)	Proliferation of bundles of smooth muscle fibers and blood vessels (Fig. 1)/Smooth muscle actin +	Angioleiomyoma	None/×1 mth no recurrence
2	8	F	Multiple nontender swellings × 3 yrs., now increasing in size/right helix, concha and preauricular/skin tags, congenital sinuses/cysts	Multiple pale and brown soft elongated cords and linear pieces and firm nodules (5 to 13)	Tortuous swollen myxoid nerves and nodules of proliferating spindle cells involving skin, fat, muscle and cartilage (Fig. 1)/S-100 protein +	Neurofibromas and plexiform neurofibroma	Café au lait macules/×1 mth no recurrence
3	23	M	On & off earache × 1 yr, now hearing loss and canal blockage × 1 mth, pedunculated bony swelling/right canal/osteoma	A gray white to yellow hard bony polyp (10)	A polypoid nodule composed of mature lamellar bone trabeculae with bone marrow spaces, and covered by skin squamous epithelium (Fig. 2)	Osteoma	Psoriasis/×4 yrs. no recurrence
4	60	F	Asymptomatic soft swelling × 2 yrs./right earlobe/lipoma	Solid round fatty lobulated nodule (10)	A lobulated nodule of mature adipose tissue	Lipoma	None/×10 yrs. no recurrence
5	23	M	Painless nodule × 1 mth/right auricle/aural polyp	A gray white to pink polypoid lobulated hemorrhagic firm nodule (8)	A polyp composed of lobules of proliferating capillary-sized blood vessels (Fig. 2)	Lobular capillary hemangioma	None/×8 yrs. no recurrence
6	19	F	Painless swelling × 3 wks/right canal/aural polyp	A yellow to brown solid firm nodule (9)	A dermal nodule composed of mononuclear foamy histiocytes and multinucleated giant cells (Fig. 2)/ CD68 +; CD1a, FXIIIa and S100 -	Juvenile xantho-granuloma	None/No follow up
7	59	M	Asymptomatic nodule × 2 yrs./right pinna/skin tag	A rubbery to firm gray white polypoid nodule (9)	A polyp composed of a non-capsulated dermal nodule of proliferating wavy spindle cells (Fig. 3)/ S100 +; SMA and FXIIa -	Neurofibroma	None/×10 yrs. no recurrence
8	25	F	Asymptomatic nodule × 2 mths/right canal/ papilloma, aural polyp	A firm to hard gray white nodule (5)	A dermal nodule composed of hypocellular hyalinized collagen bundles with clefts and storiform pattern (Fig. 3)	Storiform collagenoma (sclerosing fibroma)	None/×11 yrs. no recurrence
9	61	M	Multiple small papules × 1 mth/left helix/ previous history of ear basal cell carcinoma × 1 yr, patient worried about recurrence	Skin ellipse with multiple papules and small nodules (2 to 5)	Multiple non-capsulated dermal nodules of spindle cell proliferation (Fig. 3)/S100+	Neurofibromas	None/No follow up
10	14	M	Asymptomatic nodule × 1.5 mth/left auricle; tragus-preauricular swelling/lipoma, cyst, lymph node	A skin ellipse with a deep brown round firm nodule (7)	A well-circumscribed solid subcutaneous nodule composed of interlacing bundles of spindle cells (Fig. 4)/SMA +; CD68, FXIIIa, CD34 and S100 -	Leiomyoma	None/×4mths no recurrence
11	51	M	Asymptomatic nodule × few yrs./right pinna/cyst	A firm yellow brown round nodule (6)	A lobulated nodule composed of vascular channels (Fig. 4)/CD31 +; S100, DP-40, desmin -	Arteriovenous hemangioma	None/×3 mths no recurrence
12	17	F	Canal wax blockage × 2 mths, bony swelling/ right canal/osteoma	A gray white to yellow hard bony nodule (5)	A nodule composed of mature lamellar bone trabeculae with bone marrow spaces	Osteoma	Psoriasis, psoriatic arthritis/×7 mth no recurrence
13	60	M	Painless nodule/right canal entrance/cyst, aural polyp	A yellow brown nodule (7)	A dermal nodule composed of mononuclear foamy xanthomatous cells with frequent multinucleated Touton giant cells (Fig. 2)/CD68 +; CD1a, FXIIa, CD34 and S100 –	Juvenile xantho-granuloma: adult-type	None/×6 yrs. no recurrence
14	17	F	On & off earache and hearing loss × 1 yr/pedicle/ right canal/osteoma	A gray white to yellow hard bony nodule (5)	A nodule composed of mature lamellar bone trabeculae with bone marrow spaces	Osteoma	Acanthosis nigricans/×3 mths no recurrence

SN: serial number, M: male, mth: month, F: female, yr: year, +: positive, −: negative,

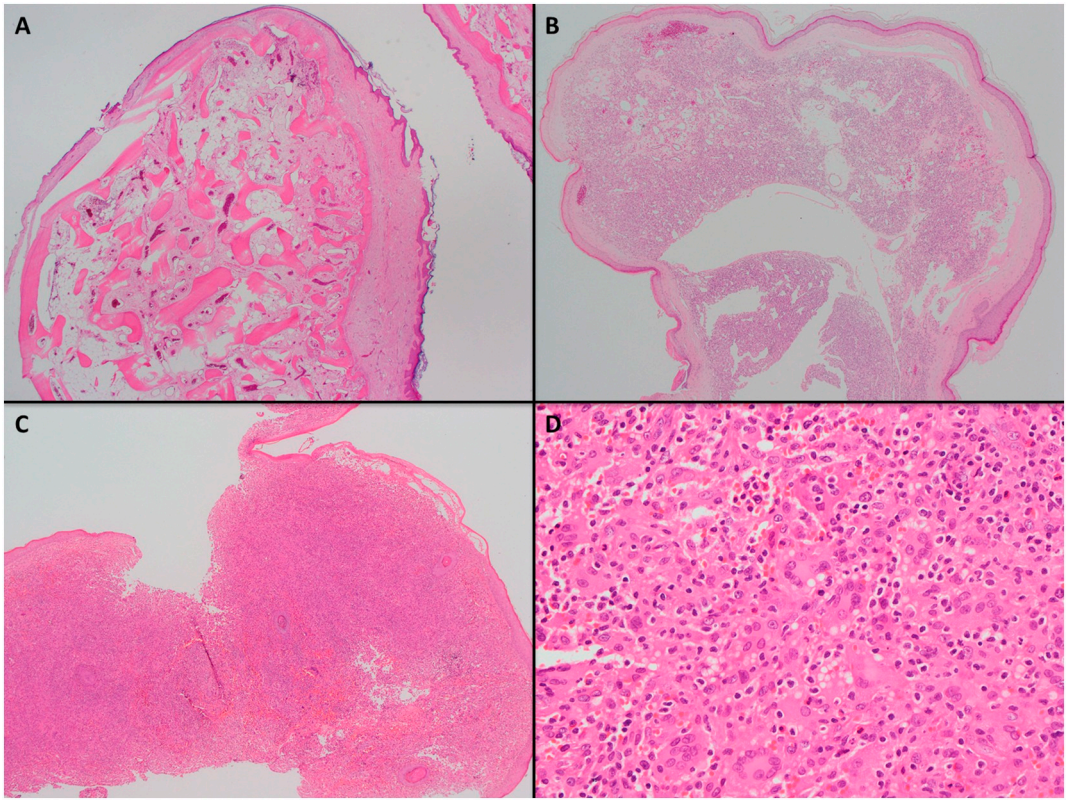

Figure 2. A) Osteoma is represented by a polypoid nodule composed of mature lamellar bone trabeculae and intervening fibrovascular and fatty bone marrow spaces (H&E ×20). B) Lobular capillary hemangioma consists of a polypoid dermal nodule composed of lobules of proliferating capillaries (H&E ×40). C) Juvenile xanthogranuloma reveals a dermal nodule composed of solid sheets of eosinophilic cells with entrapped skin appendages (H&E ×40). D) The nodule consists of plump stromal cells, foamy histiocytes and multinucleated Touton, foreign body and Langhans-type giant cells. The background showed mixed inflammatory infiltrate composed of small lymphocytes, plasma cells, eosinophils and few neutrophils. (H&E ×400).

arts are covered by skin in close proximity to perichondrium and periosteum with little if any subcutaneous tissue. They contain skin appendages, blood vessels, nerves and little muscle. Despite being a relatively small simple anatomical structure, the outer ear is made of different stromal and skeletal connective tissues derived from the mesenchyme surrounding branchial epithelia during embryologic development [5,6]. It could be considered as a unique compacted extremity. This might explain that a wide variety of lesions to affect the ear, the predilection of certain tumors and the rarity of other tumors to this site, and syndrome association of certain ear lesions [5,6]. Unfortunately, pathologists have a low tendency to consider the external ear of a special interest. The external ear may present diagnostic problems for clinicians which require the skills of histopathologists for correct interpretation. This entails familiarity with these lesions [6]. Most of the documented cases of benign stromal tumors of the external auditory canal, auricle and lobe in the English literature were single case reports or a small series of cases (Table 2). They encompass a variety of tumors some are common but rarely involve the external ear while others are generally rare tumors. They include bony (exostosis, osteoma), cartilaginous (chondroma), vascular (hemangioma), fibroblastic (fibroma), fibrohistiocytic (dermatofibroma), peripheral nerve sheath tumors (schwannoma, neurofibroma), muscular (leiomyoma) adipocytic (lipoma) tumors and myxoma. They are not exclusive to and not primarily confined to the external ear. Collectively, meatal osteomas and auricular angioleiomyomas are not infrequent tumors with a possible predilection to the external ear [7-18]. Our study demonstrated that most of the tumors can histologically be diagnosed without difficulties. Few cases, however, may impose some difficulties in differentiating them from lesions with similar overlapping morphologic features, in which, attention to certain histomorphologic features and the use of a small panel of IHC markers are helpful.

Osteomas are the most common benign tumors of the external ear with a restriction to the canal [7-12]. Clinically they are distinct from exostoses. They are solitary unilateral pedunculated polypoid nodules. Multiple osteomas might be associated with Gardener syndrome. Solitary osteomas are nonsyndromic. Whether the finding of osteomas in young patients with skin lesions, in our series, is a coincidence or an association needs further evaluation by future studies. The second most common benign tumor is cutaneous angioleiomyoma (AML) with a predilection to the auricle [13-18]. Contrary to classic AMLs, auricular AMLs are painless and do not show a female predominance. The mechanism of pain in AML is probably due to mechanical stretching or chemical stimulation by mast cells of the intralesional or capsular nerves [19]. In our case, CD117 showed frequent intralesional mast cells, but S-100 protein failed to demonstrate intralesional or capsular nerves. Whether this observation can explain why head and neck AMLs are not painful in contrast to classical AMLs needs validation by comparative studies. Variants of AMLs include cavernous, venous and solid capillary, and a transitional form that might simulate glomus tumor and hemangiopericytoma. Nonvascular deep leiomyomas usually affect extremities. Few involved the ear. They might mimic schwannoma and dermatofibroma due to presence of palisading, storiform growth pattern, circumscription, degenerative and inflammatory changes [20,21]. They are positive for smooth muscle markers. Most of the peripheral n-

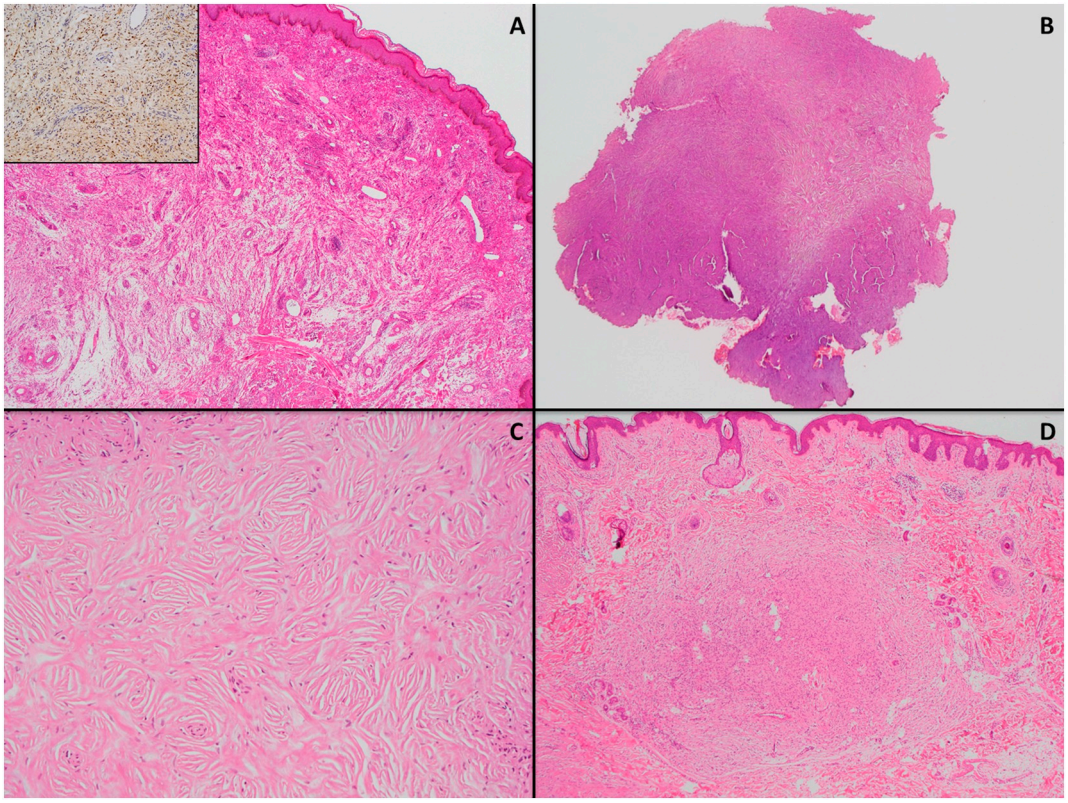

Fig. 3. A) A neurofibroma manifests as a dermal nodule composed of loosely arranged wavy spindle cells associated with blood vessels. (H&E × 40). Inset: The spindle cells are positive for S-100 protein. B) Sclerosing fibroma shows a solid fibrosclerotic nodule (H&E ×40). C) The nodule shows hypocellular eosinophilic storiform collagen fibers with characteristic clefts (H&E ×200). D) A neurofibroma as an example of the multiple superficial round dermal spindle cell nodules. The overlying epidermis is unremarkable (H&E ×40).

erve sheath tumors of the external ear are neurofibromas [22,23]. Schwannoma and perineurioma are rare [4,24]. Neurofibromas can present as multiple small superficial dermal papules or nodules, a large deep solitary nodule, or as diffuse plexiform tortuous enlarged peripheral nerves associated neurofibromatosis type 1. Small dermal NFs can be confused with dermatofibromas while plexiform myxoid NFs with myxomas. They are positive for S-100 protein. Xanthogranulomas encompass a group of histiocytic lesions. Few cases of juvenile xanthogranulomas involved the ear in young and old patients [25-28]. They should be distinguished from benign fibrous histiocytoma (BFH) especially the cellular foamy histiocytic variant which can occur in the ear [29,30]. They lack the grenz zone, epidermal changes, storiform growth pattern and hemosiderin. They show characteristic Touton-type giant cells, prominent vasculature and mixed inflammatory cells of lymphocytes, eosinophils and neutrophils. The cells are positive for CD68, CD45, CD4, and negative for FXIIIa and CD34. They can be confused with Langerhans cell histiocytosis especially when frequent eosinophils are present. They are negative for CD1a, S-100 protein and CD207. Vascular lesions of the external ear are rare [31,32]. Sclerosing fibroma, also called storiform collagenoma, is a relatively peculiar new entity [33,34]. It is a dermal nodule which can be sporadic or associated with Cowden disease. It has a characteristic hypocellular collagenous storiform clefted growth pattern. There is no previous report of ear sclerotic fibroma in the literature. Even though the external ear is rich in cartilage and to some extent adipose tissue represented in the ear lobe, chondroid and adipocytic tumors are the least common [35-37]. Clinically, the majority of benign mesenchymal ear tumors are isolated. Some tumors, particularly when multiple or recurrent might be a first hint to a hidden syndrome. Clinicians therefore should be vigilant of certain syndromic ear tumors, for example myxomas, neurofibromas, osteomas and sclerosing fibroma [6,38].

In conclusion, benign mesenchymal neoplasms of the external ear are uncommon and miscellaneous. The majority are solitary isolated lesions. Clinicians should be aware of certain tumors associated with syndromes. Meatal osteomas and auricular angioleiomyomas compared to other tumors are not infrequent with some predilection to the external ear. Histologic diagnosis is easy in most of the cases. Some may impose diagnostic challenges that warrant confirmatory immunohistochemistry. Because they can be clinically confused with other lesions, histologic examination is mandatory. This entails familiarity of pathologists with the broad spectrum of external ear disorders.

Financial and funding disclosure

No sources or grants of financial support from any institution to be disclosed. No conflict of interests to be disclaimed or financial disclosure to be declared.

Funding

This study was not funded.

Ethical approval

This article does not contain any studies with human participants. As a retrospective review, informed consent was not obtained.

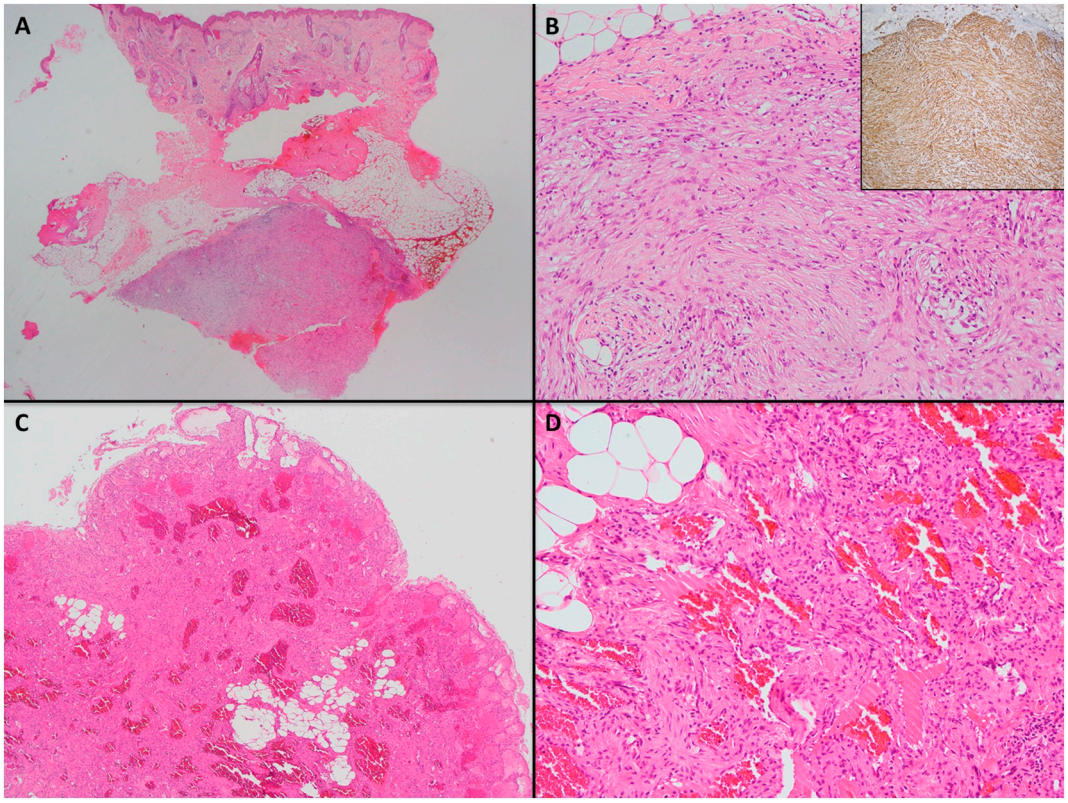

Fig. 4. A) Leiomyoma shows a deep well-defined solid subcutaneous nodule (H&E × 40). B) The nodule is composed of intersecting and storiform fascicles of plump spindle cells. The background showed mixed inflammatory cells and no muscular blood vessels (H&E ×200). Inset: The spindle cells are positive for smooth muscle actin. C) Arteriovenous hemangioma reveals a lobulated well-defined nodule composed of vascular channels of variable sizes (H&E ×40). D) The nodule consists of intermixed venous and arterial sized muscular blood vessels with entrapped adipose and fibrous tissue (H&E ×200).

Table 2
A summary table of published English literature of case series of benign mesenchymal tumors of the external auditory canal.

Author/Year	Type and number ( ) of tumors
Graham [8]/1979	Osteoma (3), exostoses (2)
DiBartolomeo [39]./1979	Exostoses (70)
Ferreiro & Carney [38]./1994	Myxoma (18)
Fenton et al. [9]/1996	Osteoma (5), exostoses (8)
Redaelli de Zinis et al. [40]/2007	Hemangioma (2)
Tanigawa et al. [37]/2008	Chondroma (5)
Hoshino et al. [41]/2012	Myxoma (2)
Yamamoto et al. [42]/2015	Hemangioma (3)
Yamahara et al. [43]/2018	Chondroma (2)

Declaration of Competing Interest

No conflicts of interest to be declared by authors.

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