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Nodular fasciitis of the external auditory canal: Clinical case report and review of the literature

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ABSTRACT

Nodular fasciitis is a benign reactive soft tissue tumor arising from fibroblasts and myofibroblasts. Its incidence is low and misdiagnosis is frequent especially for malignant lesions. This can lead to inappropriate and unnecessary invasive treatment. Nodular fasciitis of the external auditory canal is extremely rare. So far, around fifteen cases have been reported. We present here the case of a 90-year-old patient with nodular fasciitis of the right external auditory canal. The lesion extends anteriorly for 6.5 cm and reaches the posterior wall of the maxillary sinus. To our knowledge, this is the first case in the literature of an external auditory canal nodular fasciitis presenting as an inflammatory ear polyp with such a wide extension.

Contents

1. Case

A 90-year-old man showed up to an ENT consultation for spontaneous right otalgia which was exacerbated while chewing. The patient had a history of renal aneurysm, colic adenocarcinoma treated 27 years ago by chemotherapy and surgery, currently in a cured state, and an active myelodysplastic syndrome. The ENT clinical examination showed an inflammatory and fleshy polyp in the right external auditory canal (EAC), the eardrum was not visible (Fig. 1). Biopsies were performed, bringing back inflammatory material including numerous polymorphonuclear neutrophils for which no conclusion could be made. The patient was seen again in order to perform new biopsies and to perform a Computed Tomography of the temporal bone. The latter showed a full middle ear, an irregular and micro-erosive aspect of the external auditory canal as well as a focal transfixion of the hypotympanum on its external side towards the temporomandibular joint. A significant tumefaction of the soft tissues of nodular aspect was also objectified. (Fig. 2).

The anatomopathological result on a histological and immunohistochemical basis turned out to be positive for nodular fasciitis. (Fig. 3 and Fig. 4).

A Magnetic Resonance Imaging (MRI) was also performed, unfortunately of poor quality due to patient movements during the examination but it was still able to describe the size and the extension of the lesion (Fig. 5). The latter is 65 × 60 mm in size and

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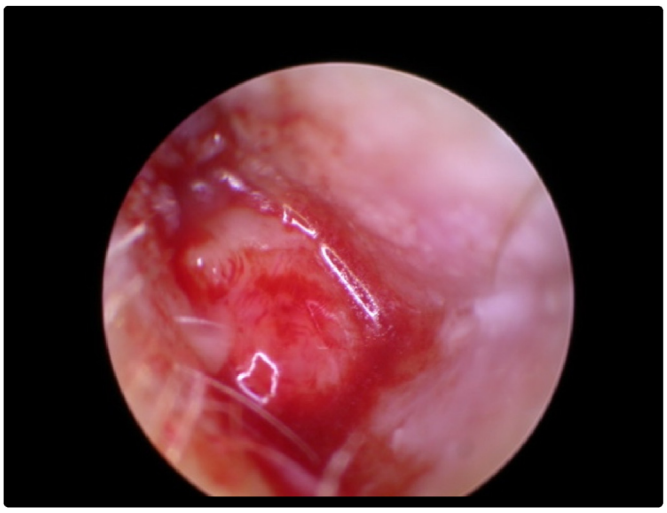


Fig. 1. Right ear microscope otoscopy.

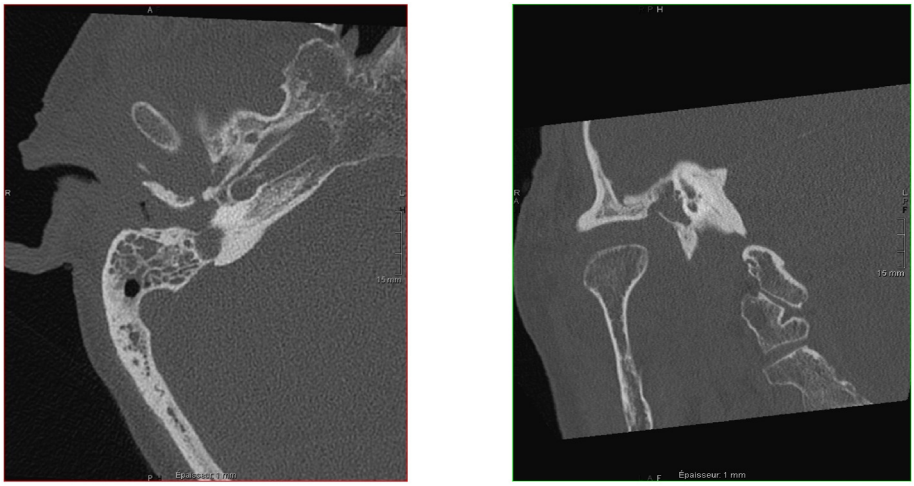
extends from the external auditory canal to the posterior wall of the maxillary sinus in the axial plane.

Given the extension of the lesion and the patient's age, empirical treatment was initiated with daily methylprednisolone 32 mg orally associated with topical treatment combining Dexamethasone and Chloramphenicol in the form of ear drops after a multidisciplinary consultation.

Even though the patient had severe hypoacusis and was wearing hearing aids, he reported an improvement of his hearing at the follow-up appointment after 18 days of treatment. However, the lesion had not changed significantly on the otoscopy. Another MRI had been scheduled but the patient died of a stroke unexpectedly before his appointment.

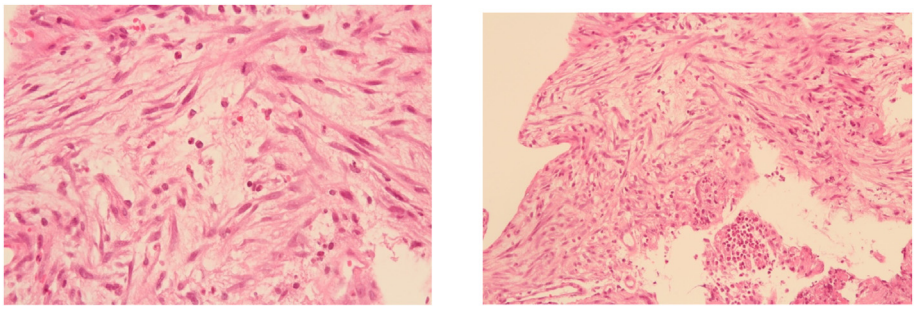
2. Discussion

Nodular fasciitis (NF) is a benign reactive proliferation of myofibroblasts, first described in 1955 by Konwaler as pseudosarcomatous fasciitis (Konwaller et al., 1955). NF has also been referred to



A B

Fig. 2. CT-Scanner Siemens Somatom Definition AS. Ultra high-resolution spiral acquisition in 0.4 mm and MPR reconstruction 1 mm: axial (A) and sagittal (B) section of the right petrous bone.



A B

Fig. 3. Histological images with hematoxylin and eosin magnification ×400 (Fig. 3A) and ×200 (Fig. 3B) demonstrating variable cellularity as well as a variable extracellular matrix, ranging from myxoid to fibrous. Note the presence of fusiform cells with a fascicular or storiform arrangement, ovoid nuclei with fine chromatin and low mitotic activity. No significant atypia. Presence of inflammatory elements, mainly scattered lymphocytes.

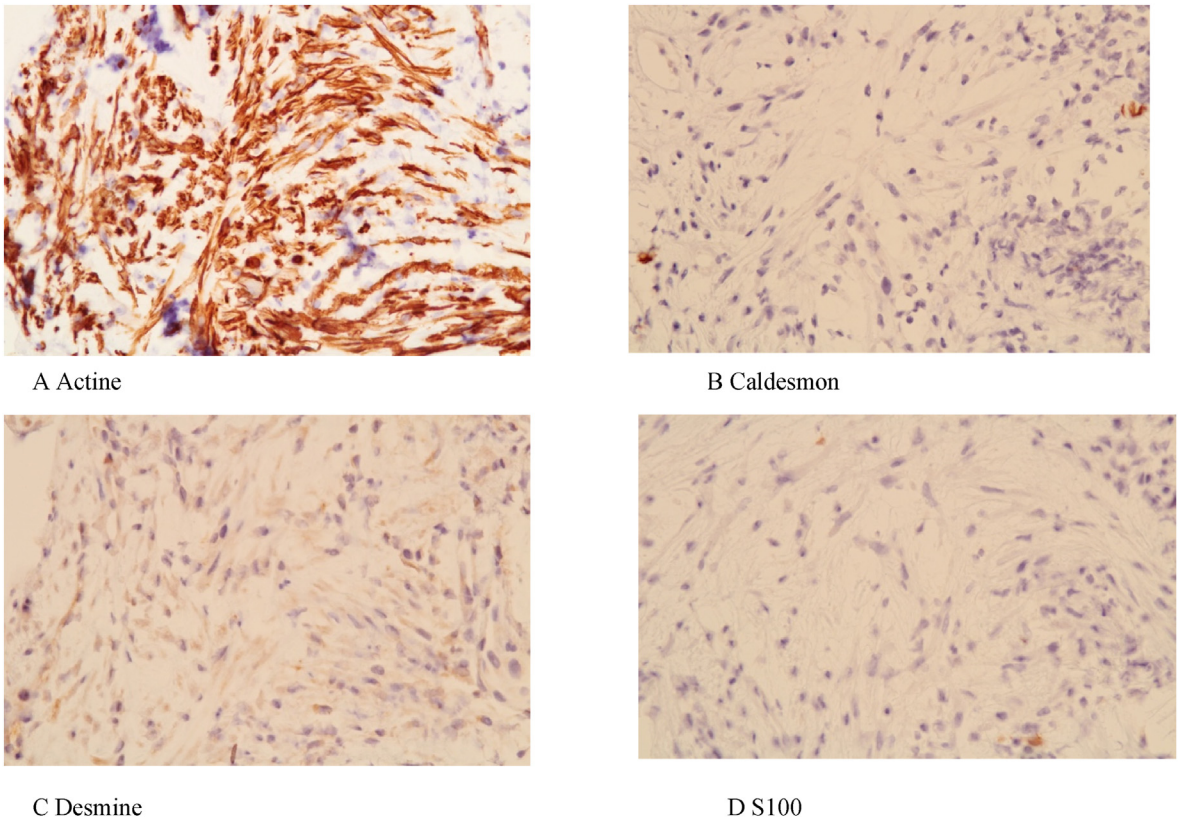


Fig. 4. Immunohistochemistry. Myofibroblastic cells are Actin positive (4.A) and negative for Caldesmon (4.B), Desmin (4.C) and protein S100 (4.D).

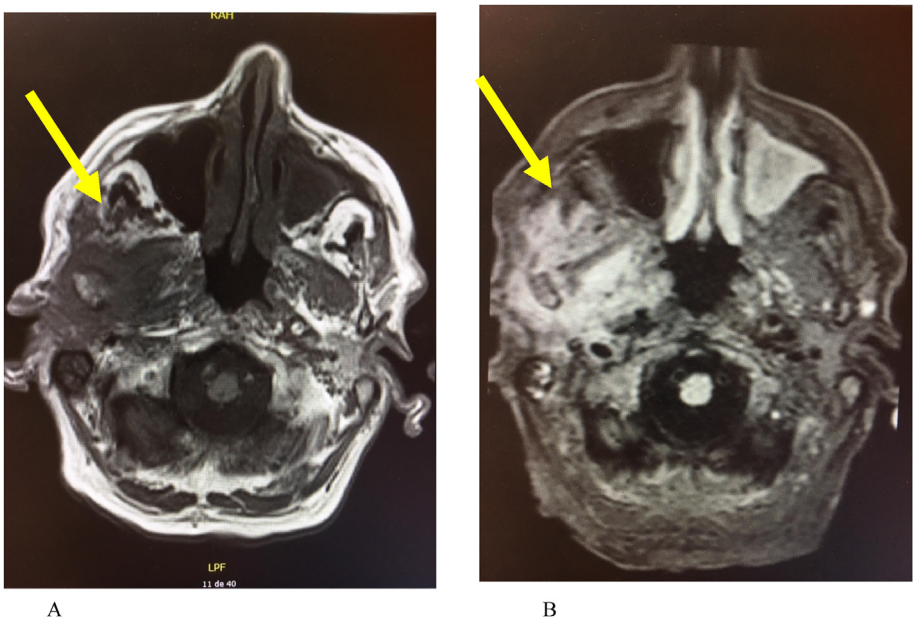


Fig. 5. MRI Siemens Magnetom Avento fit 1.5 T. Acquisition. Axial slices through the inferior turbinate in T1-weighted (Fig. 5A) and T2-weighted Flair Fat Sat (Fig5.B). The arrow pointing to the lesion: we note a filling of the right external ear and the middle ear extending by transfixion towards the temporomandibular joint with extensive infiltration of fatty and muscular tissues over 65 mm wide and of 60 mm on the anteroposterior axis around the ascending branch of the mandible. The lesion affects the deep plane of the tonsillar region and joins the posterior wall of the right maxillary sinus. It is contiguous with the right intrapetrous internal carotid and the right internal jugular gulf.

as nodular fibrositis, subcutaneous fibromatosis or proliferative fasciitis. It develops intramuscularly, from subcutaneous tissue or fascia. The aetiology remains unknown to date, although there is a notion of trauma in 15% of the cases (Weinreb et al., 2009a).

Considered as reactive lesion for a long time, recent studies have shown a genetic fusion of the MYH9-USP6 genes in 83–92% of cases

leading to the overexpression of the protein USP6 (Ubiquitin-specific peptidase 6), pro-oncogene, and which can help confirm the diagnosis (Erickson-Johnson et al., 2011; Chen et al., 2014).

Nodular fasciitis can occur throughout the human body but it is mainly located on the extremities of the upper limbs. The second most affected region is the head and neck region in the order of 3.1–24% (Konwaller et al., 1955; Meister et al., 1978; Lu et al., 2015; Dahl and karlstedt, 1980) which in this case preferentially reaches the cervical region (53%) (Lu et al., 2015). Head and neck involvement appears to be more common in children (Hseu et al., 2015; Bemrich-Stolz et al., 2010; DiNardo et al., 1991). The NF affects men more frequently (1.4 men/1 woman) with an average age of 36 years (Lu et al., 2015). It has the characteristics of a rapidly growing lesion that can reach 2–3 cm in a few weeks and can reach up to 8 cm in total (Nakagawa et al., 1994; Kumar et al., 1997). The lesions size is smaller in the head and neck region with an average of 1 cm (Weinreb et al., 2009b). It can cause mild pain or tenderness or cause symptoms through mass effect. The lesion is usually mobile and well-circumscribed.

Radiologically, NF generally appears as a well-defined hypo or isoechoic lesion on ultrasound (Lu et al., 2015). Fine needle punctures led in several cases to a wrong diagnosis of pleomorphic adenoma (Mardi et al.,2005; Al-Rahbi et al., 2015; Jain et al., 2008). Computed tomography (CT) can show a heterogeneous lesion in case of cystic or mucoid transformation with sometimes even peripheral enhancement. Usually, the FN is rather homogeneous and seems hypo-or even isodense compared to the surrounding muscles (Wang et al., 2002; Chin et al., 2003). On MRI, nodular fasciitis appears homogeneous hypointense or isointense on T1, heterogeneous and iso- or even hyperintense on T2, with enhancement after administration of gadolinium in 40% of cases (Chin et al., 2003; Dinauer et al., 2007). Specific MRI signs such as the “fascia tail sign” or the “cloud sign” can help to make the diagnosis of nodular fasciitis in soft tissue tumors (Wu et al., 2020).

Some authors suggest that nodular fasciitis should be part of the differential diagnosis of head and neck masses found superficially and showing iconographic enhancement after injection, both on MRI and CT. This might be even more relevant if there exists a history of trauma and the lesion grows rapidly (Kim et al., 2005). In addition to rapid growth, bone erosion or destruction can be observed on imaging (which can make iconographic differential diagnosis complicated) (Chin et al., 2003). Some case reports also report hyper-metabolic activity of nodular fasciitis at Fluorodeoxyglucose Positron Emission Tomography (FDG-PET) (Kessels et al., 2004; Gotthardt et al., 2010; Seo et al., 2017).

The treatment of choice is surgical excision (Bernstein and Lattes, 1982). Recurrence is often linked to incomplete excision and is low, around 1–2%. It must also suggest another diagnosis. Some case reports report a good response to the injection of intralesional corticosteroids (Hseu et al., 2015; Graham, 1999; Nishi et al., 2006). Spontaneous regressions after biopsy or incomplete excision have also been described (Nishi et al., 2006; Dahl and. Nodularfasciitis, 1981). Lesions of the face for which the excision can leave unsightly scars can therefore possibly be treated non-surgically thanks to intralesional injections of corticosteroids, provided that there is histopathological certainty of the lesion (Oh et al., 2015).

Nodular fasciitis seems to be expressed in the external ear in approximately 1.5% of cases and is the culprit for 1.9% of external ear lesions (including malignant, benign or inflammatory lesions) according to a case series of 50 cases of external ear nodular fasciitis (50 out of 2930 auricular lesions) (Lu et al., 2015; Lester et al., 2001). At this location, they are usually smaller (average 1.9 cm) and recurrences are more frequent (9.3%) (Agarwal et al., 2013). Among nodular fasciitis of the outer ear, 12% are located in the external auditory canal (6 out of 50), making it a fairly rare region of presentation. In this case series of 50 cases of external ear nodular fasciitis, a wrong diagnosis was made in ¾ of the cases. It should be noted that 30% (9 cases out of 30) were wrongly considered as sarcomas after pathological analysis. This is why NF is also called “pseudosarcomatous fasciitis” because it has often been confused with a sarcomatous lesion due to its similar pathological characteristics (mitotic activity, cellularity and infiltration) associated with its clinical rapid growth. In these cases, a wrong diagnosis is likely to lead to more aggressive treatment with significant side effects. Therefore, it is imperative for physicians (clinicians and pathologists) to recognize nodular fasciitis presentation and characteristics.

To our knowledge, this case is the first presentation of nodular fasciitis as an inflammatory ear polyp with such extension (Table 1). Ear polyps can be due to inflammatory pathologies (infectious, cholesteatomatous, granulomatous) or neoplasms (benign or malignant) (Tay and Hussain, 1997). The infectious origin could be eliminated from negative bacteriological samples, a normal blood test (except a slight increase of the C-reactive protein) and the absence of purulent secretions. Moreover, the patient did not have predisposing conditions to malignant otitis externa such as diabetes or chronic renal failure. Polyps are associated with cholesteatomas in 25–45% of cases but these polyps usually arise from the attic or from posterosuperior marginal tympanic membrane defects. The absence of squamous epithelium with a granular cell layer or keratine on biopsies and the iconographic extension were not in favor of this type of pathologies. Benign tumors may also present with an ear polyp such as a jugular glomus tumor, middle ear adenoma, or facial nerve neuroma (Tay and Hussain, 1997). However, the data from the imaging performed on our patient did not point in this direction. The large size of the lesion and the associated osteolysis raised suspicion of an invasive and malignant lesion. Langheransian histiocytosis is an extensive osteolytic pathology that can cause an ear polyp. However, it usually occurs in young people and nearly half of the cases present bilateral involvement as well as pathological sites other than the temporal bone (Modest et al., 2016). The typical histological appearance showing multinucleated Langerhans cells, eosinophils and histiocytes was also not found on biopsies.

Malignant tumors of the external auditory canal or middle ear are rare. Squamous cell carcinomas are the most common followed by adenoid cystic carcinomas and basal cell carcinomas. A malignant ear polyp might be either a primary lesion or the result of the extension of adjacent tumor sites like the parotide, the temporomandibular joint, the infratemporal fossa, nasopharyngeal, the pre ou retroauricular region. Malignant melanoma, squamous cell carcinoma, adenocarcinoma, cystic adenoid carcinoma and soft tissue sarcomas of the ear and temporal bone might present with ear polpys. Anatomopathology analysis-clarification was needed and as previously mentioned, differentiating NF from sarcomas might be a difficult task for the contributing pathologist. Soft tissue sarcomas of the ear are more common in children and approximately 50% of these patients have neurological signs at the time of diagnosis (Kuhel et al., 1996). Despite the significant iconographic extension our patient showed no cranial nerve impairment.

In the majority of cases, ear polyps are simple benign inflammatory polyps (Agarwal et al., 2013). External auditory canal lesions are usually small and localized, so diagnosis is often delayed. An effective biopsy is also a challenge, leading in many cases to inadequate biopsies which are too superficial or scattered. Therefore, it is recommended to proceed to a biopsy and/or wide local excision with a rim of healthy tissue. Repetition of the procedure is suggested in the case of unfavourable evolution or doubtful and/or non-contributory histopathological results.

Table 1
MEDLINE/Pubmed literature review of external ear canal nodular fasciitis.

	Age	Ear trauma	Exact localisation	size	symptoms	treatment	Time of follow-up	recurrence
Abdel-Aziz et al. (2008): 6 cases	mean age of 7 years and 3 months	none	No information	0.5–1.5 cm	- All cases: unilateral earache - 3 cases: bleeding associated with offensive discharge in two of them	Complete surgical removal	One year	2 cases: - In one case after two months - In one case after 4 months. No recurrence after second chirugical removal
Halsey et al. (2020): case report	19 months	Increase of an ear lesion after trauma	Concho bowl with obstruction of the external auditory canal	2.1 × 2.1 × 0.8 cm	Ulceration and bleeding	Complete surgical removal	No information	No recurrence
Ahn et al. (2016): case report	19 years old	none	Posterior wall of the cartilaginous portion of the right external auditory canal	1.7 cm	itching	Complete surgical removal	One year	No recurrence
Liu et al. (2021): case report	17 months	No information	Cavity of the auricular concha	1 cm	none	Complete surgical removal	One year	No recurrence
Milo et al. (1999): case report	39 years old	No information	Lesion involving the left external auditory canal and middle ear cavity with extension inferiorly to the parotid gland, temporomandibular joint, masseter, and pterygoid muscles	No information	minimal pain and drainage	Complete surgical removal	13 months	No recurrence
Lester et al. (2001): 6 cases of EAC nodular fasciitis among 50 cases of auricular nodular fasciitis	no detailed subgroup information	3 out of 6	no detailed subgroup information	1.3 cm (mean size)	no detailed subgroup information	surgical removal (no detailed subgroup information)	From 0.4 to 29.0 years (median, 11.4 years) with no detailed subgroup information	Yes (1 out of 6)
Della Volpe et al. (2022): case report	4 years old	none	Lesion involving the left tragus and the lateral portion of the external auditory canal	1.7 × 2.1 × 2.4 cm	No symptoms at the first meeting	Complete surgical removal	One year	No recurrence
Wang et al. (2022): 1 case of EAC nodular fasciitis among 3 cases of auricular nodular fasciitis	1year and 9 months	none	Left ear with no other information	1.7 × 1.6 × 0.9 cm	Painless lesion with bleeding	Complete surgical removal	8 months	No recurrence
Yver (2021): case report	49 years old	No information	Left concha bowl	2 × 1.5 cm	No information	Complete surgical removal	No information	No recurrence
Cobanoglu et al. (2010) : case report	8 years old	Yes, 3 weeks earlier	Cartilaginous part of the inferior canal wall of the left external auditory canal	1.7 × 1.0 × 1 cm	No information	Excisional biopsy with positive surgical margins	7 months	The lesion recurred within a month, no recurrence 7 months after a total excision.

Declaration of competing interest

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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