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SCIENTIFIC LETTER

Solitary fibrous tumor in the thyroid gland as an incidental finding

Tumor fibroso solitario en la glándula tiroides como hallazgo incidental

A 62-year-old woman was referred to the Endocrinology clinic in September 2022 due to the incidental detection of a multinodular goiter on a supra-aortic trunk ultrasound requested by Ophthalmology for a central retinal vein thrombosis.

During the index visit to the Endocrinology clinic, the thyroid ultrasound performed revealed the presence of a bilobar multinodular goiter with multiple cystic and mixed nodules up to 10 mm in diameter (ACR TIRADS 1 and 2), along with a poorly defined dominant lesion of nearly 4 cm in the middle third of the right lobe, showing intense hypoechogenicity without internal vascularization (ACR TIRADS 4). This nearly anechoic nodule did not exhibit posterior acoustic enhancement, and after considering it a possibly cystic lesion, an attempt was made to aspirate it using fine-needle aspiration biopsy (FNAB) in the clinic. Very little hemorrhagic content was obtained, and the cytological result was Bethesda I, non-diagnostic. The patient was referred to Otolaryngology for a right hemithyroidectomy, which turned out uneventful (Fig. 1A).

The pathological description of the lesion reported histological and immunohistochemical findings consistent with a solitary fibrous tumor without histological features of aggressiveness. Tumor size was 40 mm × 32 mm × 31 mm (Fig. 1B). Histology revealed the presence of proliferation of spindle cells with elongated nuclei, fine chromatin, blunt edges, scant pleomorphism, and nuclear atypia; cells were arranged in interlaced bundles, with areas of a storiform pattern and abundant intervening collagenous stroma. Cellular density was described as mild to moderate, with a mitotic index of 3 mitoses per 10 high-power fields, no evidence of necrosis, and clear surgical margins (Fig. 1C, D). Immunohistochemistry turned out positive for CD34 and STAT6 (Fig. 1E, F) and negative for the remaining requested markers: S100, calcitonin, desmin, chromogranin, AME, AML, CK19, EMA, and CK19.

Solitary fibrous tumors (SFTs) are fibroblastic lesions with a ubiquitous location, affecting adults between 20 and 70 years of age[1]. The age-adjusted incidence is 0.61 cases per million person-years for extrameningeal SFTs and 0.37 for meningeal SFTs.[1] Among extrameningeal SFTs, the most frequent locations are the abdominal cavity (31%), limbs (29%), pleura (22%), trunk (11%), head and neck (7%).[1]

SFTs of the thyroid gland are very rare, with few cases described. Taccagni et al. were the first to study them in 1993, describing 3 cases of thyroid SFT with an age of onset between 32−61 years.[2] Among the most recent and extensive reviews is that by Zhang et al., which describes a total of 43 cases collected between 1993 and 2022[3]. The patients' mean age was 54.5 years (19 women and 24 men); the mean tumor diameter, 58 mm, and the most frequent clinical presentation was a slow-growing, painless cervical mass.[3] In a later review, Santoro et al. collected up to 48 cases.[4] Regarding diagnosis, there are no characteristic ultrasound findings for these tumors, and it can be challenging due to the low yield of cytological material obtained via FNAB, often due to fibrosis or paucicellular areas of the tumor.[4] In these cases, core needle biopsy (CNB) may be useful, as it allows for obtaining more thyroid tissue and enables immunohistochemical techniques. Ha et al. demonstrated in 85 nodules with highly suspicious ultrasound images previously reported as benign on FNAB that CNB could reveal malignancy, and that cases with benign histology had abundant fibrosis justifying their intense hypoechogenicity, similar to SFTs.[5]

Histological findings are characteristic and consist of a proliferation of spindle cells with elongated nuclei and scant cytoplasm, arranged in a storiform or patternless pattern, on a variable collagenous stroma with interspersed dilated and branched staghorn capillaries.[1,6] Immunohistochemically, they are notable for strong and diffuse positive staining for CD34 in more than 80% of SFTs, although this is a nonspecific marker that may be lost in malignant SFTs.[1] Additionally, nuclear expression of STAT6 is characteristic, with this marker being highly specific (>85%) and sensitive (98%) for SFTs, making it the most reliable marker for diagnosis.[1,6] Other markers with variable and more nonspecific expression include CD99, bcl-2, and vimentin.[1,6] The pathogenesis of these tumors is based on the fusion of NAB2-STAT6 genes, which act as transcription activators, causing dysregulation of the early growth response (EGR) signaling pathway.[1]

The patient was presented to a multidisciplinary committee, which decided not to extend the surgery to total thyroidectomy given the absence of histological features of malignancy. The prognosis of SFT is calculated using risk prediction models for metastasis, such as that of Demicco et al. from 2017.[7] This model categorizes the risk of metastasis in

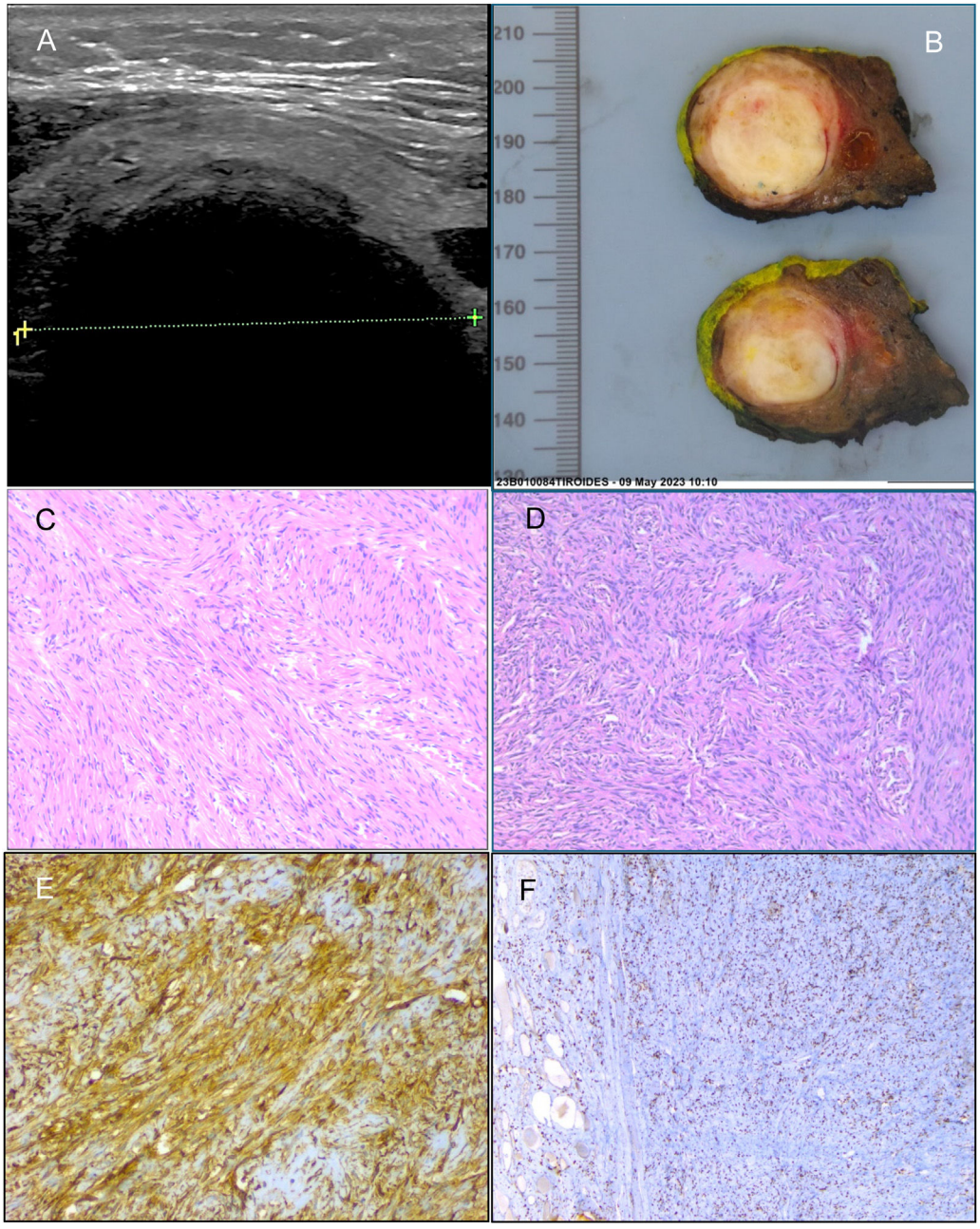

Figure 1 Ultrasound (A), macroscopic (B), histological (C, D), and immunohistochemical (E, F) images of solitary fibrous tumor. Ultrasound showed an anechoic nodule without posterior acoustic enhancement (A). On macroscopic section, a well-demarcated, whitish, firm nodular lesion measuring 4 cm in maximum diameter was identified (B). Microscopically, a proliferation of spindle cells with a fascicular (C) and whorled (D) growth pattern was noted. In the immunohistochemical study, tumor cells tested positive for CD34 (E) and STAT6 (F).

SFTs into low, intermediate, or high based on 4 different variables: age, tumor size, mitotic index, and percentage of necrosis[7]. Our patient scored 2 out of 7, indicating a low risk of metastasis, so chronic follow-up was chosen.

Long-term postoperative follow-up for SFTs is mandatory due to their unpredictable biological activity.[8] The follow-up recommended by the National Comprehensive Cancer Network (NCCN) for these types of types of tumors without metastasis is every 3–6 months for the first 3 years and then annually.[9] The cervicothoracic computed tomography scan and thyroid ultrasound performed in our patient 6 months and 1 year after the intervention, respectively, showed no local recurrence. In the review by Santoro et al., only 2 out of 48 thyroid SFTs had local recurrence or distant metastasis.[4] However, of note, in our hospital, another case of thyroid SFT was reported in a 46-year-old man with no prior conditions who presented with liver and cervical metastases 4 months after diagnosis, indicating that the risk of dissemination is not negligible.[10] This patient did have histopathological markers of poor prognosis, with a high

mitotic index of 8 mitoses per 10 high-power fields and a Ki-67 of 30%.[10]

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