

A rare localization of primary osteosarcoma is presented. A woman aged 76 years was operated on for rapid growth of thyroid right lobe tumour. Histopathology showed anaplastic cancer with numerous foci of osseous metaplasia, negative with thyroglobulin, calcitonin, synaptophysin and chromogranin. A high proliferative activity of the tumour was observed (MIB-1 reaction) in the form of a positive reaction in approx. 40% of the tumour cell nuclei. The tumour stage was evaluated as pT4aNxMx according to the TNM scale. The reconsultation revealed negative staining with cytokeratin, and positive with vimentin, thereby confirming the mesenchymal origin of the tumour, with the final diagnosis being primary thyroid osteosarcoma. Taking into consideration the histopathological diagnosis, the extremely low radiation sensitivity of the tumour, the patient's age, the radical surgical treatment and persisting respiratory failure, radiotherapy was rejected in favour of further follow-up. The patient remains under oncological and endocrinological care.

**Key words:** thyroid gland, tumour, osteosarcoma, pathology.

## Primary osteosarcoma of the thyroid gland – a case report

Ewa Zembala-Nożyńska, Dariusz Lange

Department of Tumour Pathology, Maria Skłodowska-Curie Memorial Cancer Centre and Institute of Oncology, Gliwice Branch, Poland

### Introduction

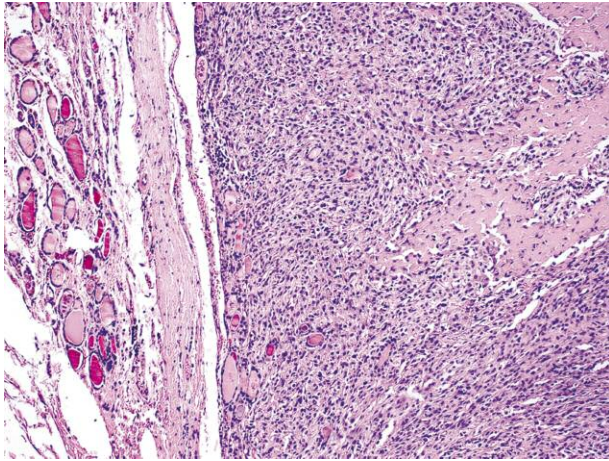
Osteosarcoma constitutes a small percentage of rare malignant non-epithelial neoplasms of the thyroid, and has been reported in only several dozen cases [1]. It mimics anaplastic carcinoma with extensive osseous metaplasia [2, 3]. One of the conclusive tests is immunohistochemical staining, which excludes epithelial differentiation of the tumour [4]. We present a case of a female patient who underwent surgery due to a fast-growing tumour of the right thyroid lobe, with an unexpected final diagnosis.

### Case report

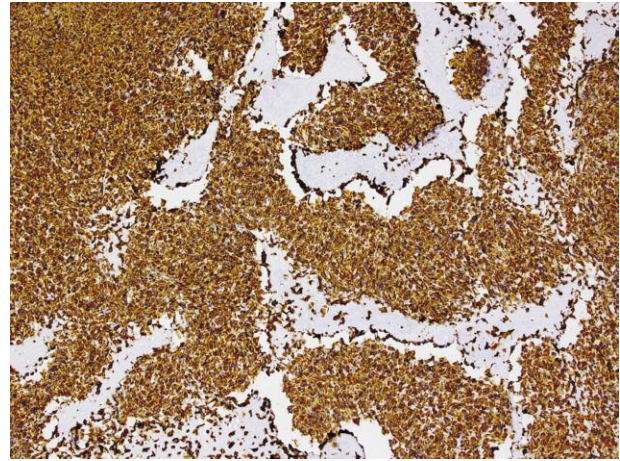
A female patient, aged 76, was qualified for surgical removal of a tumour of the right thyroid lobe, within the substernal goitre, due to its rapid growth over a period of two months. Computed tomography revealed nodular goitre with massive calcifications, reaching the sternal notch. The estimated size of the goitre, measured in cross-section through both lobes, was 9 × 6 cm. The trachea was significantly displaced to the left and narrowed. No qualifying biopsy was carried out before surgery, opting for radical strumectomy.

With an incision at the base of the neck, having cut through the short muscles, the thyroid lobes were identified. The left lobe was the size of a plum, with nodular reconstruction, whilst the right lobe was the size of a large fist, expanding down the lower pole, past the sternum and pushing the trachea to the left (approx. 4 cm off the central line). Both lobes were hard, fused with the trachea, with little mobility. The muscles were separated and the upper and lower poles were ligated. Having freed the lateral parts of the lobes of the thyroid from the trachea, the small pyramidal lobe was removed. The gland was removed in its entirety and two Redon drains were inserted. The muscles, subcutaneous tissue and the skin were sutured. The material was sent for histopathological examination. The early post-operative period was complicated by acute respiratory failure requiring intensive care treatment. The impairment of vocal cord mobility resulted in the necessity to insert a tracheotomy tube on the laryngology ward.

The evaluation of the single-piece surgical material revealed a nodular tissue structure, with an irregular external surface and dimensions of 9 × 6 × 7.5 cm, without separated thyroid lobes. A correct thyroid texture was observed only in the left lobe, with a diameter of approx. 2.5 cm. The remaining part of the material, corresponding to the right thyroid lobe of a diameter of approx. 7 cm, was a greyish-white tumour of increased density, with extensive calcified foci. The histopathological examination showed anaplastic carcinoma with numerous foci of osseous metaplasia. Immunohistochemical staining showed negative reactions with thyroglobulin, calcitonin, synaptophysin and chromogranin. A high proliferative activity of the tumour was observed (MIB-1 reaction) in the form of a positive reaction in approx. 40% of the tumour cell nuclei. The tumour stage was evaluated as pT4aNxMx according to the TNM scale.



**Fig. 1.** Fragment of thyroid tumour with osteoid formation. Staining H&E, magnification 100×



**Fig. 2.** Thyroid tumour stained immunohistochemically for vimentin. All cells are positively stained. DAB, magnification 100×

A month after the operation, in quite good general condition, the patient reported for oncological treatment. The postoperative scar at the base of the neck was fully healed, with no infiltration. The peripheral lymph nodes were slightly enlarged, with small oedema of the subcutaneous tissue. Computed tomography with local staging was carried out with a view to qualification for radiotherapy. A histopathological reconsultation of the tumour was requested.

The reconsultation revealed negative staining with cytokeratin, and positive with vimentin, thereby confirming the mesenchymal origin of the tumour, with the final diagnosis being primary thyroid osteosarcoma.

Taking into consideration the histopathological diagnosis, the extremely low radiation sensitivity of the tumour, the patient's age, the radical surgical treatment and persisting respiratory failure, radiotherapy was rejected in favour of further follow-up. The patient remains under oncological and endocrinological care.

## Discussion

Osteosarcoma, the most frequent malignant bone tumour, may also occur in various organs outside the osteoarticular system. However, these are rare incidents, described in single cases and involving the heart, lung, prostate, urinary bladder and breasts [1–4]. The extrasosseous form of osteosarcoma occurs very rarely (approx. 5%), especially in the thyroid, where, so far, it has been diagnosed in 28 cases [5], and in only two cases with fine-needle biopsy [6, 7]. This tumour is characterised by fast growth and requires quick surgical intervention due to pressure-related symptoms of dyspnoea.

The clinical course mimics anaplastic thyroid carcinoma [8]. Beside sarcomatoid poorly differentiated thyroid carcinoma [6] and anaplastic carcinoma with osseous metaplasia, the differentiating diagnosis should additionally involve medullary thyroid carcinoma, spindle epithelial tumour with thymus-like differentiation (SETTLE) [9], synovial sarcoma and metastasis of osteosarcoma [10].

The tumour is characterised by a variety of histological images, from extensive monomorphic infiltrations of primary mesenchymal cells with high mitotic activity, in which it is

generally difficult to observe osteoplasia, through spindle-cell and epithelioid areas with accompanying giant tumour cells with multiple nuclei. The presence of bone matrix and mature bone trabeculae is a frequent component of the tumour. The images of fine-needle biopsy are particularly challenging diagnostically. Cytological material is usually rich in cells and appears in compact tissue fragments with slight discohesion, frequently showing a high degree of cell polymorphism. There are spindle cells and epithelioid cells with varying amounts of cytoplasm, from trace to abundant, frequently with the presence of intracytoplasmic vacuoles. The nuclei of these cells are usually elongated or oval-shaped, with coarse-grained chromatin and quite distinct chromocentres, often eccentric as in plasmocytes. In the background, there are usually binuclear and polynuclear cells with considerable polymorphism, some of which resemble osteoclasts [6, 7]. Beside cells, there may be masses partially composed of fibrillar structures, adjacent to tumour cells. Thus, the cytological image may be ambiguous and requires histopathological confirmation, supported by immunohistochemical examinations. Immunohistochemical examinations with vimentin (positive reaction), cytokeratin, TTF1, calcitonin, chromogranin, synaptophysin and S-100 (negative staining) are of decisive significance.

The clinical course is characterised by rapid progression, which, within weeks, leads to death in the mechanism of lung metastasis or the superior vena cava syndrome as a result of extensive infiltration of the perithyroid tissue and large carotid vessels. Treatment consists in radical strumectomy and radiotherapy, with supplementary chemotherapy.

## References

1. Trowell JE, Arkel DG. Osteosarcoma of the thyroid gland. *J Pathol* 1975; 119: 123-7.
2. von Sandersleben J, Hänichen T. Tumours of the thyroid gland. *Bull Wld Hlth Org* 1974; 50: 35-42.
3. Syrjänen KJ. An osteogenic sarcoma of the thyroid gland (report of a case and survey of the literature). *Neoplasma* 1979; 26: 623-8.
4. Hertel V, Basten O, Bockmühl U. Extrasosäres Osteosarkom der Schilddrüse. *Laryngorhinootologie* 2006; 85: 913-6.
5. Makis W, Novales-Diaz JA, Hickeson M. Primary thyroid osteosarcoma: staging and evaluation of response to therapy with F-18 FDG PET-CT. *Clin Nucl Med* 2010; 35: 640-3.

6. Tong GX, Hamele-Bena D, Liu JC, Horst B, Remotti F. Fine-needle aspiration biopsy of primary osteosarcoma of the thyroid: report of a case and review of the literature. *Diagn Cytopathol* 2008; 36: 589-94.
7. Syrjänen KJ. Fine needle aspiration cytology of the thyroid osteosarcoma. Report of a case. *J Cancer Res Clin Oncol* 1980; 96: 319-23.
8. Nitzsche EU, Seeger LL, Klosa B, Freudenberg N, Moser EA. Primary osteosarcoma of the thyroid gland. *Nucl Med* 1992; 33: 1399-401.
9. Lee FY, Wen MC, Jan YJ, Wang J. The predominant monophasic variant of spindle epithelial tumour with thymus-like differentiation (SET-TLE) of neck soft tissue with late pulmonary metastasis. *Pathology* 2010; 42: 188-90.
10. Lam KY, Lo CY. Metastatic tumors of the thyroid gland: a study of 79 cases in Chinese patients. *Arch Pathol Lab Med* 1998; 122: 37-41.

**Address for correspondence**

**Ewa Zembala-Nożyńska**  
Wybrzeże Armii Krajowej 15  
44-101 Gliwice, Poland  
e-mail: jerno@wp.pl

**Submitted:** 3.11.2011

**Accepted:** 8.12.2011