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CASE REPORT

HETEROTOPIC OSSIFICATION IN DIFFUSE SCLEROSING PAPILLARY THYROID CARCINOMA- A RARE VARIANT WITH RARER METAPLASIA IN UNUSUAL AGE: CASE REPORT

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ABSTRACT

Background: Squamous metaplasia may be diagnosed in some diffuse sclerosing variants of papillary carcinoma. Objective- However, bone maturing in the thyroid tumour is a phenomenon that occurs rarely and can mimic a variety of lesions. This study reports such a case in 83 year old man presenting with painless thyroid swelling from last 5 years. **Methods**: Case study. **Conclusions:** Osseus metaplasia in diffuse sclerosing variant can be considered as a different subtype because of its aggressive behaviour.

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INTRODUCTION

Thyroid diseases form a major bulk of endocrine disorders in India. Thyroid cancer is increasing, ranking it as the ninth most common cancer worldwide[1] with Projected incidence rate 1 in 285 women and 1 in 759 men.[2] Papillary carcinoma being the commonest. However, diffuse sclerosing (DSV-PTC) variant is rare. Thyroid cancer nodules can undergo degenerative changes such as cystic, hemorrhagic and fibrotic changes and in rare cases can calcify or ossify. Heterotopic ossification (HO) or osseous metaplasia (OM) is transformation of soft tissue into bone. However, OM with mature bone formation rarely occurs. Here we report a case of DSV-PTC with squamous metaplasia (SM) and HO with mature bone formation

CASE REPORT

An 83 year old man came to OPD with the complaint of painless neck mass from the last five years, with no significant past history.

Tests for serum anti thyroglobulin and antisomal antibodies were positive. Thyroid function tests showed that the patient was euthyroid. On the basis of these physical and laboratory findings, initial impression was either Hashimoto's thyroiditis or thyroid cancer. The neck ultrasound reported a hypoechoic TIRADS 4b nodule in the left lobe. FNA was performed. The aspiration sample provided a hypercellular smear of pleomorphic cells. Numerous pseudo nuclear inclusions and few longitudinal grooves were identified (Fig 1). No colloid was seen in this sample. BETHESDA V compatible with suspicion of malignancy was given. Total thyroidectomy was decided with central compartment and left level III lymphadenectomy. Gross examination showed diffuse replacement of parenchyma by cystic space surrounded by white firm tissue and measuring 3.5x1.8x1.5cm, which was gritty on cutting. Histopathology showed marked cystic degeneration lined by tufts of hobnailed neoplastic cells with pseudoinclusions, numerous psammoma bodies and lymphocytic inflammation (Fig 2). Extensive foci of squamous differentiation were seen. The stroma showed calcification along with dense stromal fibrosis and sclerosis.

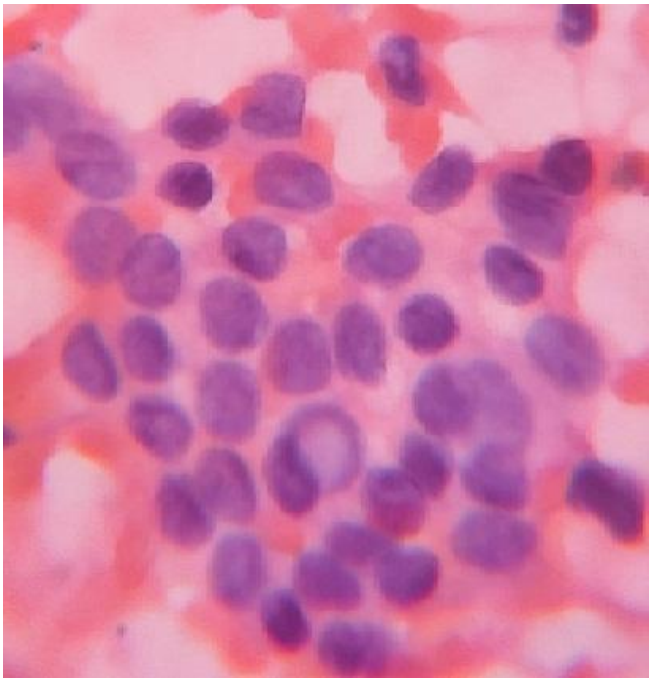

Fig 1. Photomicrograph of FNA smear showing nuclear groove and pseudoinclusion (H&E1000x)

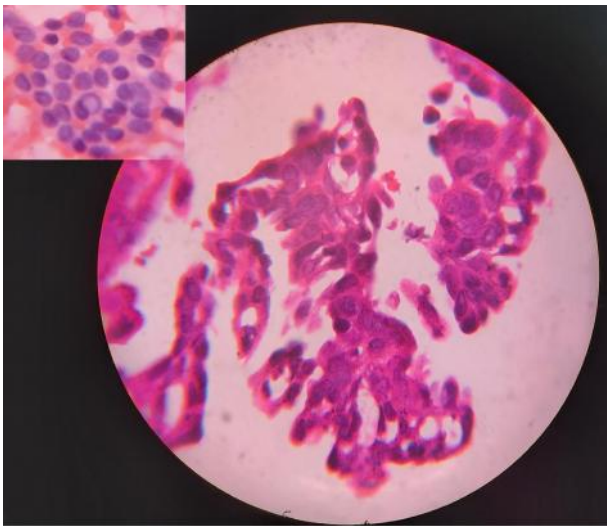

Fig 2. Photomicrograph of surgical specimen displaying papillae with nuclear features (H&E400x). [Inset- Fig 1]

Occasional foci displaying beginning of ossification from this stromal calcification were noted (Fig 3). Sclerotic tissue was seen intervening mature bony trabeculae. Extra thyroid extension was not evident. Central compartment lymph nodes showed cystic change grossly and microscopically with metastatic deposits Classified as pT3N1.

DISCUSSION

The incidence of this variant among all the papillary carcinomas of the thyroid is low, but this is not a rare condition if the patient is a child or young woman. Our patient is an elderly man. Typical histologic features include-

Diffuse involvement of one or both lobes
Fibrosis
Heavy lymphoplasmacytic infiltrate
Abundant psammoma bodies

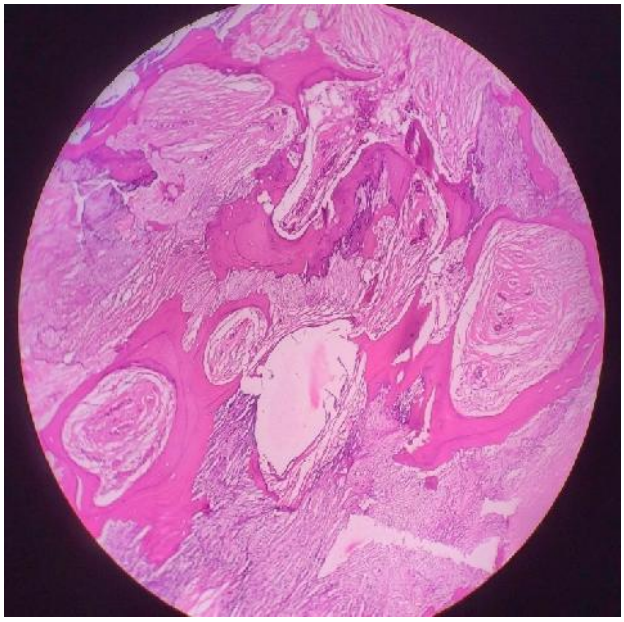

Fig 3: Photomicrograph showing lamellar bone, extensive fibrosis, dystrophic calcification and squamous morules (H&E400x)

SM can be seen. DSV-PTC can display not only characteristic psammoma bodies, but also calcification or/and bone metaplasia. However, this is an incidental and rare finding. Rupture of follicles often results in granulomatous inflammation with fibrosis which further leads to calcification and ossification.[3] Different theories have been proposed to explain the pathogenesis of this phenomenon but none has effectively elucidated the exact mechanism.

One theory suggets upregulation of Bone Morphogenetic Protein (BMP), osteonectin, osteocalcin and osteopontin in tumor cells causing metaplasia of the pleuripotent stromal cells. BMP 1 (metalloproteinase) activates procollagen I, II,III and VII forming extracellular matrix.[4] Calcified tissue has more BMP2 which along with basic fibroblast growth factor (bFGF) signals cellular proliferation.[5] The prognostic significance of DSV-PTC remains debatable. This variant is associted with extrathyroid extension and higher rate of local and distant metastases (usually in the lungs) at presentation and shorter disease free survival compared with classic PTC. Never theless, the overall mortality rate is similar.[6] PTC with OM has not been described as a specific variant in latest WHO classification[7]because of its rarity. Further studies might define whether it is a true histological subtype and whether it has any prognostic implication.

CONCLUSION

DSV-PTC is a rare entity and very few cases have been reported with metaplastic changes. Despite the aggressive surgical management, the recurrence rate is high. However, when associated with heterotopic bone formation, it could be a more aggressive subtype that can affect the prognosis. Further studies are needed to determine whether it can be considered as a separate histological variant and to study its treatment and prognostication.

CONFLICT OF INTEREST: Nil.

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GLOSSARY OF ABBREVIATIONS

List of Abbreviations (in alphabatical order)

bFGF- Basic Fibroblast Growth Factor
BMP- Bone Morphogenetic Protein
DSV-PTC - Diffise sclerosing variant of papillary thyroid carcinoma.
HO- Heterotopic Ossification
OM- Osseous Metaplasia
SM- Squamous Metaplasia

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