MALIGNANT ECTOPIC THYROID TISSUE WITH DISTANT METASTASIS

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BACKGROUND:
Ectopic Thyroid Tissue comprises around 41-68% of the cases of thyroid dysgenesis. It is predominantly found in females, constituting 65-80% of cases. Ectopic thyroid tissue may become goitrous or may result in hypo or hyperfunctioning. Primary malignant transformation in such tissue is quite a rare entity. Although any form of malignancy can develop at such sites but follicular carcinoma is found to be the dominant form. Very infrequently, malignant ectopic thyroid tissue can present with metastasis to lymph nodes. To the best of our knowledge, distant metastasis has never been reported with primary malignant ectopic thyroid tissue. Here we report a first case of its kind of malignant ectopic thyroid tissue over manubrium sterni with distant metastasis to ilium & spine.

CASE REPORT:
A 42-year-old Pakistani female presented in Endocrine clinic at Aga Khan University, Karachi, Pakistan with complaint of gradually increasing swelling on anterior aspect of manubrium sterni for last 6 months. She had no goitre and was clinically euthyroid. Rest of systemic examination was also unremarkable. Computed Tomography (CT) chest showed a circumscribed soft tissue density mass arising from sternum, measuring 3.9 x 3.9cm, causing erosions of anterior, right lateral & posterior walls of sternum. Tructut biopsy of the sternal mass proved it to be thyroid tissue with follicular differentiation and occasional mitotic figures. Multiple nodules of variable sizes were also noted in Thyroid gland with no lymphadenopathy on ultrasound Neck. She was found to be biochemically euthyroid. She underwent total Thyroidectomy & excision of ectopic thyroid tissue over manubrium sterni. Surgeons noted no apparent physical connection between the primary thyroid gland and the tissue over manubrium sterni. Extensive histopathological examination of primary thyroid gland showed benign nodular hyperplasia with no evidence of malignancy. Ectopic thyroid tissue showed minimally invasive follicular thyroid carcinoma with tumor size of 3.2 x 2.3cm (Figure 1). Her postoperative 21-day biochemical profile showed TSH 22.345µIU/ml (0.4 to 4.2), Serum Thyroglobulin (TG) 88.3ng/ml, Anti-Thyroglobulin antibodies <20.0IU/ml. Based on the histopathological diagnosis, she underwent 30mCi Radioactive Iodine (RAI)131 ablation. The post-ablative whole body scan (WBS) showed good uptake of ablative dose over thyroid bed and evidence of multiple well-defined rounded areas of abnormally increased tracer accumulation over mid and lower dorsal spine, lumbar region and right iliac bone, representing bone metastasis (Figure 2). It was followed by Magnetic Resonance Imaging (MRI) spine which showed foci of metastatic deposits in T1-T3 and L2 vertebral bodies & abnormal signals showing post-contrast enhancement in superior mediat-inum with invasion of medial ends of both clavicles, more marked on the right side. So patient was found to have malignant ectopic thyroid tissue with distant bone metastasis. Consequently, she was planned to undergo a higher dose of RAI ablation after a few months. Five months after the surgery, her biochemical profile showed stimulated TSH 30.302µIU/ml, Serum TG 52.9ng/ml and Anti-Thyroglobulin Antibodies <20.0IU/ml. MRI Neck showed significantly reduced abnormal patchy enhancement along soft tissues around medial ends of clavicles. She underwent a second session of 150 mCi RAI131 ablation. This time, post-ablative WBS showed multiple areas of increased tracer deposition over mid-dorsal spine, upper lumbar region, sternal and the right iliac crest region (Figure 3). Patient is still under treatment by Endocrinology team and will be closely monitored for sign of any progression or regression of disease.

DISCUSSION AND CONCLUSION:
Malignancy in ectopic thyroid tissue is a rare encounter. Even if diagnosed, it has been found to metastasize locally only. To the best of our knowledge, we describe the first case of its kind having malignant ectopic thyroid with metastasis to spine & ilium. Even the site of ectopic thyroid tissue in our patient is a unique one, being the anterior aspect of manubrium sterni. The usual sites mentioned in the literature include base of tongue, heart, ascending aorta, humerus, oesophagus, duodenum, gallbladder, stomach bed, pancreas, mesentry of the small intestine, Porta Hepatis, adrenal gland, ovary, fallopian tube, uterus and vagina. As there was no local metastasis found at the time of surgery, so she was given a small dose of RAI131 ablation. Later the post-ablative WBS revealed the distant metastasis. Consequently patient had to go through another big dose of RAI131 ablation. So such cases appear difficult to manage and may impose difficulties in making their treatment decisions. This case warrants further research to recommend doing some form of imaging study (either diagnostic WBS or MRI neck + chest + abdomen) before going for surgery in patients with ectopic thyroid tissue to document the presence of any metastasis.

REFERENCES: