

14%), and other (43, 13%) organ systems. Unknown primary malignancies were diagnosed in 19 (5.8%) patients. Male predominance was observed in GU (72%) and lung (62%) metastases, but equal gender distributions were noted for all other metastases, $p=0.01$. Patients with GI and lung metastases were diagnosed with smaller tumors (median 2.1 and 2.6 cm, respectively) compared to those with GU (median 3.5 cm) and other (median 4 cm) metastases, $p=0.0008$. Bilateral metastases were more frequently found in patients with lung (37%) and other (38%) metastases compared to those with GI (17%) and GU (24%) metastases, $p=0.01$. Of 99 (30%) patients with bilateral metastases, 23% developed primary adrenal insufficiency (PAI), most commonly in those with lung (36%) and GU (30%) malignancies.

Only 123 (38%) patients were evaluated by an endocrinologist. Pheochromocytoma work-up was more often pursued if seen by an endocrinologist (71% vs. 15%, $p<0.0001$) in 118 (36%) patients. Adrenalectomy was performed in 94 (29%) patients, most frequently in those with GU metastases (57%), compared to only 10% of those with lung metastases. Patients were followed for a median time of 14 months (range 0.1–181), and 222 (68%) died. GU metastases carried the best prognosis with a mortality rate (MR) of 43%, as opposed to a MR of >70% in all other metastases, with lung metastases carrying the worst prognosis (MR of 85%). Multivariate analysis revealed that mortality was associated with increasing age (OR 1.3 (95% CI 1.04–1.6) for each decade) and metastasis subgroup (lung vs. GU: OR 7.2 (95% CI 3.7–14)).

Conclusion: Adrenal metastases most commonly originated from lung, GU and GI malignancies, with a third of patients discovered incidentally. Bilateral metastases occurred in 30% of patients, where 1 in 4 developed PAI. Only a minority were evaluated by an endocrinologist or had work up for pheochromocytoma. Mortality was highest in those with adrenal metastases originating from the lung.

Reproductive Endocrinology

MALE REPRODUCTIVE CASE REPORTS

Unusual Presentation of Aromatase Excess Syndrome

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Introduction: Aromatase excess syndrome is a rare disorder with gynecomastia being the main symptoms. Its prevalence is unknown with approximately twenty cases reported. We describe an unusual case of Aromatase excess syndrome. It was diagnosed incidentally at a much older age than expected while evaluating for hypersomnia.

Case presentation: A 28 year old male with no significant past medical history, presented with complaint of hypersomnolence, developed during puberty. He had multiple evaluations with no apparent etiology; sleep study and all his other laboratory tests were normal including testosterone levels, normal IGF-1 and cortisol. When patient was evaluated in the Endocrine clinic, he was found to have

bilateral gynecomastia, which he had for many years. His estradiol was 150 pg/ml (Normal <50 pg/ml). Repeat was 137 pg/ml with normal DHEA-S. Subsequent concomitant estradiol of 204 pg/ml with an estrone of 35.7 pg/ml (9–36). Total testosterone was normal at 588 ng/dl. Evaluation for a tumor with abdominal CT, testicular ultrasound, and HCG was negative. As his symptoms of fatigue and hypersomnolence were not improving and his estradiol to testosterone ratio was >10, he was started on an aromatase inhibitor and his ratio dropped from 1:40 to 1:24, as his estradiol went down to 75 pg/ml. **Discussion:** Gynecomastia is the benign proliferation of breast tissue due to imbalance between estrogen and testosterone. It could be caused by medications or medical illnesses. Occasionally its presence can harbor a serious endocrine issue especially if presenting in the prepubertal period. Thus, evaluation is often necessary. Among the pathological causes is the Aromatase excess syndrome. In this syndrome there are three types of cryptogenic genomic rearrangements identified. Those rearrangement affect the aromatase gene CYP19 and results in gain of function of the aromatase enzyme. Patients will have high estradiol and estrone level, lower FSH and LH levels that will normalize after treatment with aromatase inhibitor. Their testosterone levels could be low or normal. For the clinical diagnosis, there are four criteria; bilateral gynecomastia, pre or peripubertal onset, exclusion of other causes of gynecomastia and having a genetic trait. The first three criteria are indispensable for diagnosis while fourth one is not obligatory, but rather pathognomonic. An elevated estradiol to testosterone ratio above 1:10 is a supportive finding, as well as having a low FSH with low to normal LH. Genetic identification of the CYP19 A1 mutation remains the definitive method of diagnosis. Our patient met the first three criteria and also had estradiol to testosterone ratio is > 1:10. Genetic confirmation is challenging. Consequently, whole genome sequencing may be required. Though unusual, this case highlights the importance of looking deep in the differential when evaluating gynecomastia.

Bone and Mineral Metabolism

BONE AND MINERAL CASE REPORTS I

Autoimmune Polyglandular Syndrome Type 1 in a Patient with Bipolar Disorder

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Autoimmune Polyglandular Syndrome Type 1 (APS-1) is clinically defined as the presence of at least two components of the classic triad of hypoparathyroidism, adrenal insufficiency and mucocutaneous candidiasis. It is commonly seen amongst Finns, Sardinians and Iranian Jews and is a very rare condition, with a challenging set of management.

50-year old female with a known past medical history of Bipolar disorder, Primary Adrenal Insufficiency, Hypothyroidism, Alopecia was transferred from an acute psychiatric facility for medical clearance. Patient was noted to have findings initially suggestive of Subarachnoid Hemorrhage on a CT scan of the Head which was later