

Interhospital conference

14th July 2023

Identification Data

- ผู้ป่วยหญิง อายุ 56 ปี
- อาชีพแม่บ้าน
- สถานภาพ สมรส
- ภูมิลำเนา จังหวัด ปทุมธานี
- Underlying disease : Hypertension
- Current medication : Losartan(50) 1x1 po pc
- Chief complaint : ได้รับการส่งตัวจากรพ.ปทุมธานี เพื่อรักษา severe hypercalcemia

Question 1: จงซักประวัติเพิ่มเติม (F1)

Present illness

- 20 ปีก่อนมาโรงพยาบาล(อายุ 36 ปี) ให้ประวัติท้องผูกเรื้อรัง ทานยาระบายอาการไม่ดีขึ้น เคยไปตรวจส่องกล้องคัดกรองมะเร็งลำไส้ ผลปกติ
- 17 ปีก่อนมาโรงพยาบาล (อายุ 39 ปี) มาตรวจรักษาเพิ่มเติมเรื่องท้องผูกที่ โรงพยาบาลธรรมศาสตร์ ตรวจพบ hypercalcemia ได้รับการวินิจฉัย Primary hyperparathyroidism
- ได้รับการผ่าตัดแบบส่องกล้อง วันที่ 22/06/48 ตอนอายุ 39 ปี (17 ปีก่อน)
- ไม่สามารถตามประวัติเก่า, operative note, ผล LAB และ parathyroid imaging
- US KUB (9/06/48) ก่อน Surgery → multiple bright echoes with acoustic shadows in the pyramidal regions of both kidneys, suggestive of **pyramidal nephrocalcinosis**

Present illness

- ปฏิเสธประวัติไข้เรื้อรัง ไอเรื้อรัง/ไอเป็นเลือด หรือสัมผัสผู้ป่วยวัณโรค
- มีประวัติวินิจฉัย PHPT S/P endoscopic surgery ปี 2548
- ปฏิเสธประวัติฉายแสงที่คอ
- ปฏิเสธประวัติได้รับ lithium, HCTZ, vitamin A, active vitamin D, teriparatide
- ปฏิเสธการใช้ยาชุด ยาหม้อ ยาสมุนไพร
- ไม่มีหยุดหายใจง่าย ใจสั่น หรือท้องเสียถ่ายเหลว
- ปฏิเสธปัสสาวะเป็นก้อนกรวด หรือประวัตินิวไนไต ไม่มีปัสสาวะมีเลือด/กรวด/ทรายปน
- มีท้องผูก ถ่ายทุก 3-5 วันจากเดิมถ่ายทุกวัน ไม่มีท้องผูกสลับท้องเสีย ไม่มีถ่ายเป็นเลือด
- ปฏิเสธก้อนโตตามตัวบริเวณอื่น
- ปฏิเสธประวัติดื่มสุรา และสูบบุหรี่
- ปฏิเสธประวัติโรคกระดูกพรุน/โรคแคลเซียมสูง/โรคไทรอยด์/พาราไทรอยด์ภายในครอบครัว
- Menopause อายุ 54 ปี ,มีลูก 1 คน ตอนผู้ป่วยอายุ 23 ปี

Physical examination

- BT 36.5 PR 110 BP 114/60 RR 20/min SpO2 99%RA
- BW 62 kg Height 170 cm BMI 21.45
- GA : Good consciousness, no Cushingoid appearance, no acanthosis nigricans
- HEENT: no pale conjunctiva, anicteric sclera, no thyroid enlargement, no neck mass, no cervical lymphadenopathy, no surgical scar
- Breast : no palpable mass
- Heart: regular rhythm ,normal S1, S2, no murmur
- Chest: Clear and equal breath sound both lung
- Abdomen: normoactive bowel sound, soft, not tender, liver and spleen can't be palpate
- Ext: no edema, no dry skin
- Neuro: Alert, follow to command, motor power grade V all extremities

Problem List

- Female 56-year-old presented with
 - PTH dependent hypercalcemia
 - History of PHPT S/P endoscopic surgery ¶ 2548

**Question 2: สาเหตุของ PTH dependent hypercalcemia
ในผู้ป่วยรายนี้ (F2)**

Causes of PTH dependent hypercalcemia

- Recurrent or persistent primary hyperparathyroidism
 - ไม่สามารถตามประวัติ ผล lab หลังการผ่าตัด และ pathological study ได้
 - คนไข้ให้ประวัติท้องผูกดีขึ้นหลังผ่าตัด และติดตาม 10+ ปี → recurrent PHPT > persistent PHPT
- Multigland disease
 - Asynchronous parathyroid adenoma → recurrent PHPT
 - Parathyroid hyperplasia → persistent PHPT
- Parathyromatosis
- Parathyroid carcinoma

Question 3: ท่านจะส่งตรวจเพิ่มเติมอะไรบ้าง (F2)

Investigation

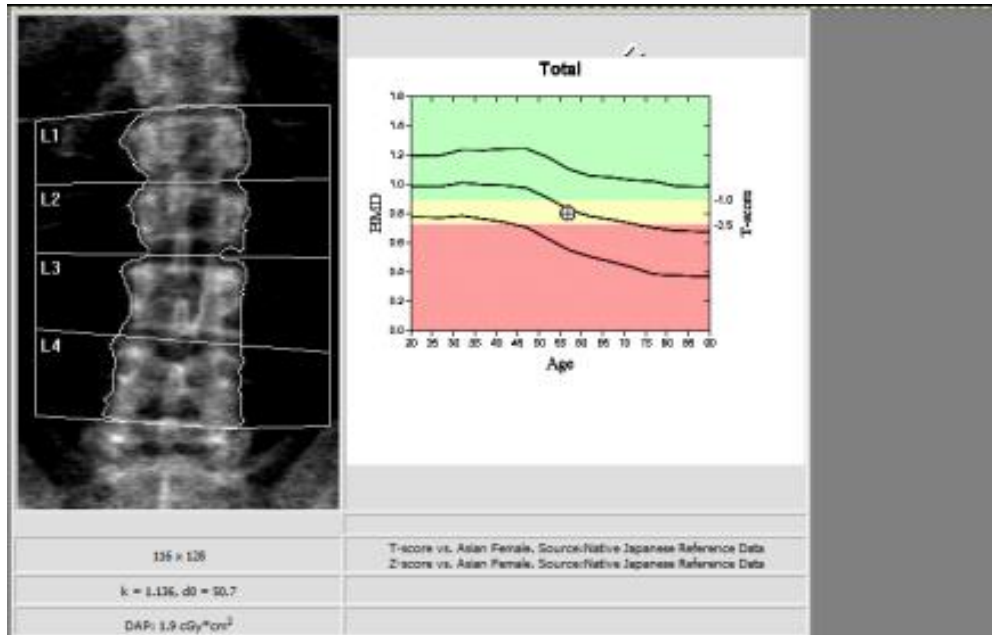
- Hb 12.3 Hct 36.5 WBC 8124 K N 53% L 34% Plt 235000
- BUN 31, Cr 1.78, Na 141, K 3.7, Cl 111, CO2 25
- Alb 3.39 Glo 2.90 TB 0.67 DB 0.16 AST 16 ALT 9 ALP 138
- FT4 1.05(0.93-1.7) FT3 2.19(2-4.4) TSH 3.04(0.27-4.2)
- EKG : NSR rate 60 , QTc 372

Investigation

1st Admission : Severe hypercalcemia

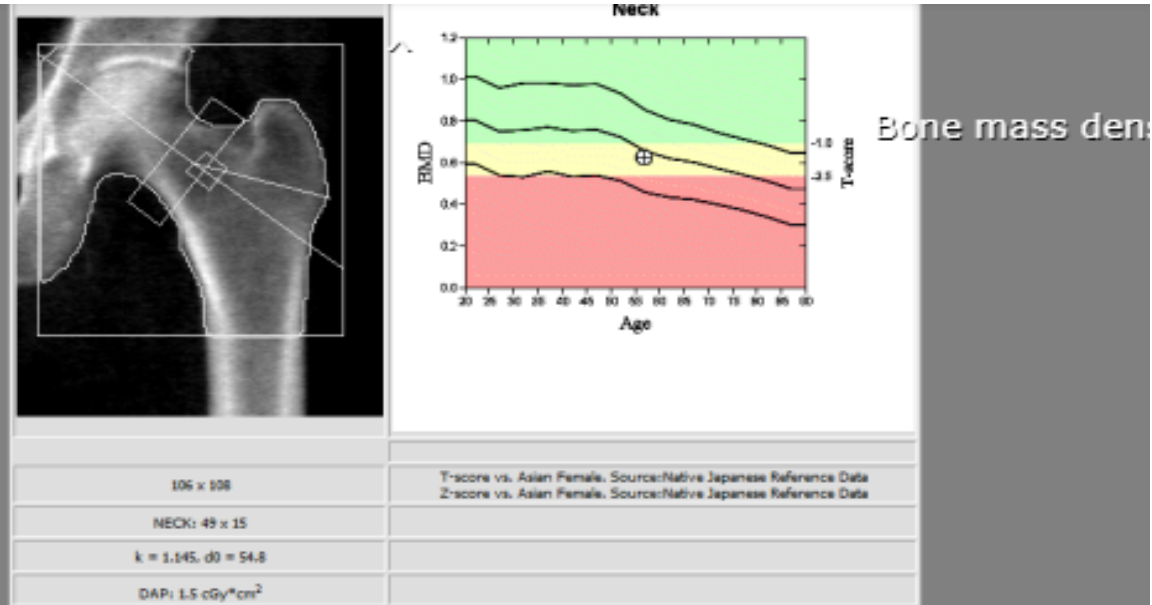
Date	Time	Ca	Alb	cCa	iCa	PO4	PTH	Vit D	Mg	Cr/GFR	Mx
14/11/65		14.3	4.39	13.9		1.9			1.3	1.98(27)	
24/11/65	14.20	14.4	4.15	14.28	8.1	2.1	1543	28.8	1.2	1.78(31)	1 st Zolendronate 4 mg iv drip in 30 min
	23.00	13.4		13.28							
25/11/65	06.00	13.2	3.38	13.6		1.7			1.9	1.53 (40)	
	15.41	12.2		12.6							
26/11/65		11.2	3.64	11.5		1.6				1.43 (43)	
27/11/65		10.3	3.39	10.7					1.5	1.43	
28/11/65		10.4	3.5	10.8		1.9				1.54	

BMD 3 parts 24/11/65



Results Summary:

Region	Area [cm²]	BMC [(g)]	BMD [g/cm²]	T-score	PR (Peak Reference)	Z-score	AM (Age Matched)
L1	13.04	10.33	0.792	-1.5	82	-0.1	98
L2	12.24	9.02	0.737	-2.1	75	-0.5	92
L3	15.33	12.86	0.838	-1.7	81	-0.1	99
L4	17.16	14.12	0.823	-1.7	80	-0.1	99
Total	57.78	46.32	0.802	-1.8	80	-0.2	97

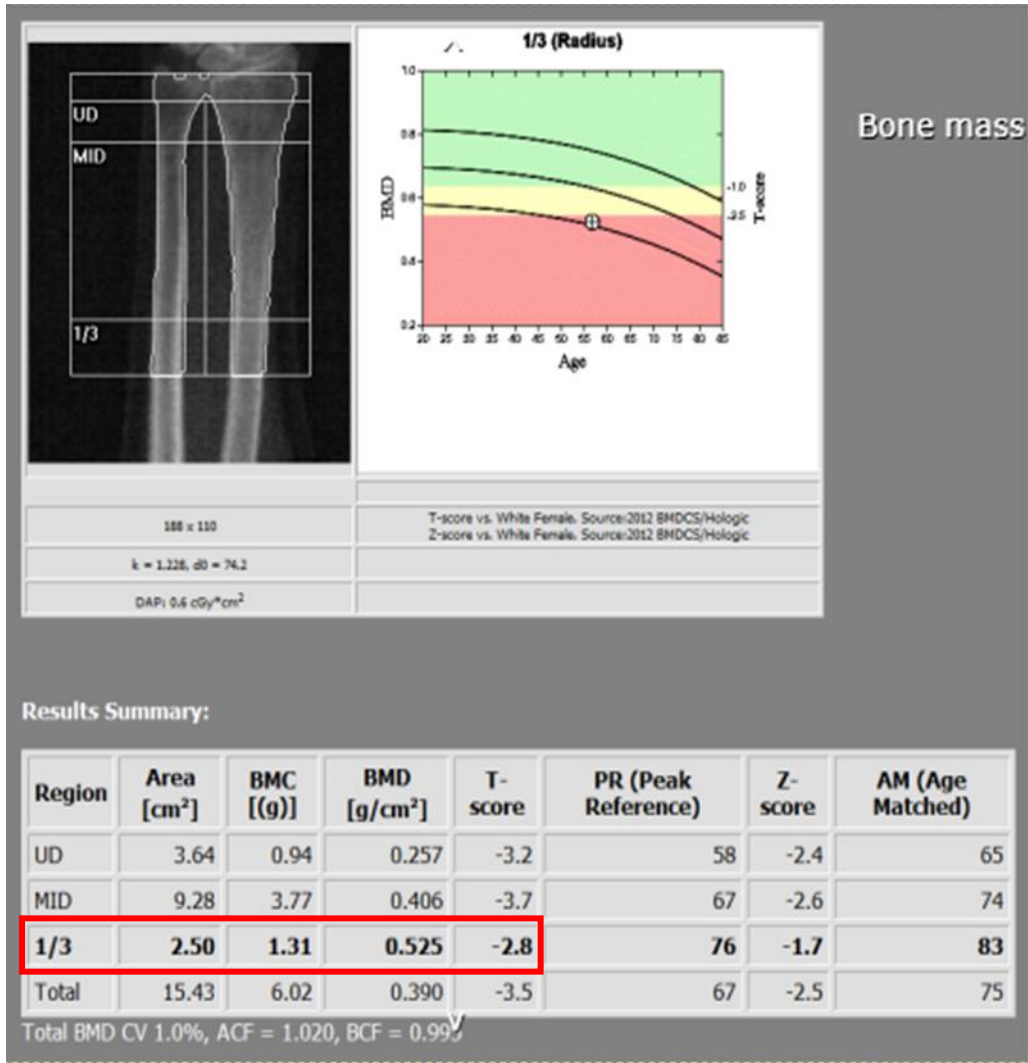


Results Summary:

Region	Area [cm²]	BMC [(g)]	BMD [g/cm²]	T-score	PR (Peak Reference)	Z-score	AM (Age Matched)
Neck	4.90	3.06	0.625	-1.7	78	-0.3	95
Troch	10.12	5.62	0.555	-0.9	87	-0.2	97
Inter	23.01	19.19	0.834	-1.2	83	-0.7	90
Total	38.04	27.88	0.733	-1.0	86	-0.4	95
Ward's	1.11	0.46	0.415	-2.4	62	-0.6	87

Total BMD CV 1.0%, ACE = 1.020, BCF = 0.995, TH = 5.211

BMD 3 parts 24/11/65



VFA 2/12/65



Vertebral Assessment

Label	Height (mm)			Percent Deformation		
	Post	Mid	Ant	Wedge	Biconcave	Crush
	Deformity (Grade)					
T4	14.6	15.3	15.5	0.0%	-4.3%	5.5%
	Normal					
T5	15.0	14.6	13.3	11.4%	2.9%	0.0%
	Normal					
T6	13.9	15.1	15.6	0.0%	-8.9%	11.2%
	Normal					
T7	13.5	14.5	15.2	0.0%	-7.6%	11.2%
	Normal					
T8	13.6	15.2	15.4	0.0%	-11.9%	11.8%
	Normal					
T9	15.3	16.6	17.2	0.0%	-9.0%	11.1%
	Normal					
T10	18.9	16.7	18.3	3.2%	11.8%	0.0%
	Normal					
T11	20.5	17.5	19.9	2.9%	14.7%	0.0%
	Normal					
T12	19.3	19.0	19.0	1.6%	1.6%	0.0%
	Normal					
L1	21.4	20.5	21.8	0.0%	3.8%	1.9%
	Normal					
L2	19.1	20.7	23.6	0.0%	-8.3%	18.9%
	Normal					
L3	18.4	20.9	22.2	0.0%	-13.8%	17.3%
	Normal					
L4	17.7	18.1	17.5	0.9%	-2.4%	0.0%
	Normal					
Std Dev	1.0	1.0	1.0	5.0%	5.0%	5.0%

* denotes automatic assessment update

Bone survey



Bone survey



Bone survey



Bone survey



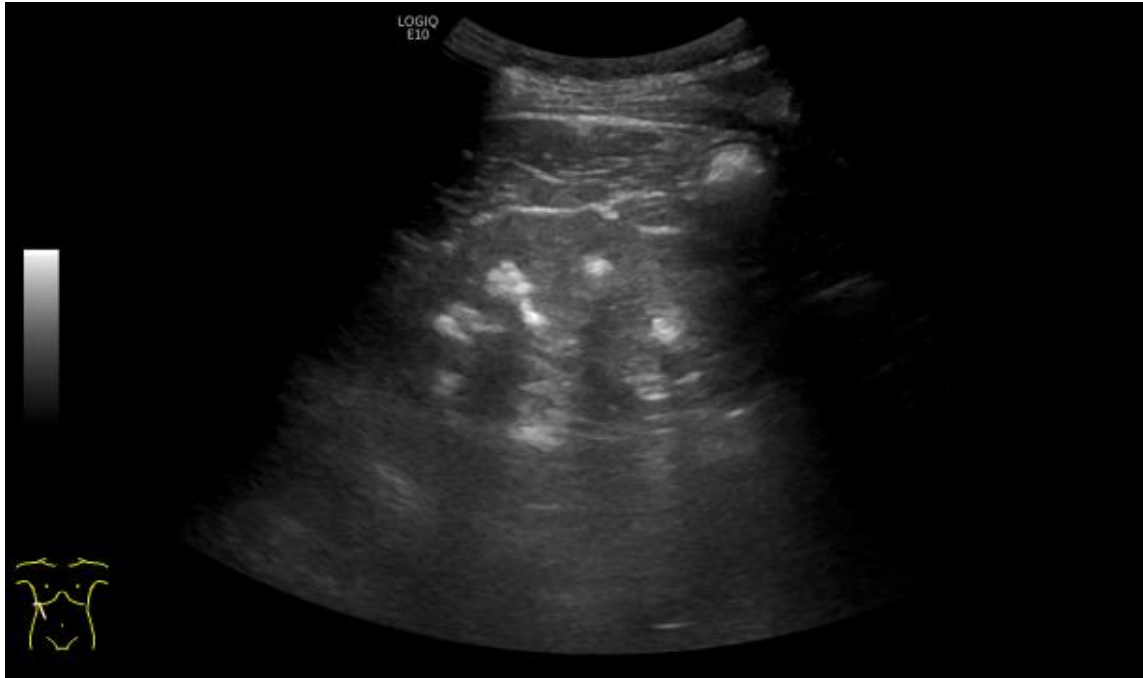
Bone survey

- **IMPRESSION:**
 - **Diffused osteopenia** of bony calvarium and T-L spine
 - No gross other osteolytic lesion
 - No gross acute fracture or dislocation
 - Mild degenerative change of thoracic spine
 - Degenerative change of lumbar spine and bilateral SI joints
 - **Medullary nephrocalcinosis** at visualized both kidneys

Film plain KUB



U/S KUB



Hyperechoic renal medullae with acoustic shadowing, representing **medullary nephrocalcinosis**

U/S parathyroid, 4DCT

	Rt upper	Rt lower	Lt upper	Lt lower	Note
US parathyroid 22/09/65		A 0.2 x 0.4 cm mix hypoechoic – hyperechoic nodule	Few 0.6 -1.4 cm nodules hypoechoic lesion Multiple 0.3 cm nodules	A 1.5x3.0 cm mixed solid-cystic hypoechoic- hyperechoic nodule at lower pole of left thyroid ddx thyroid mass, parathyroid adenoma	
4dCT	A well-defined hypodense nodule 0.4 x 0.9 x 0.9 cm Favor LN	A well-defined hypodense nodule 0.6 x 1.1 x 0.8 cm Favor parathyroid adenoma	A well-defined hypodense nodule 0.4 x 0.8 x 1.3 cm Favor parathyroid adenoma A well-defined hypodense nodule 0.5 x 0.7 x 0.7 cm Favor LN	A heterogenous hypodense nodule 2.2 x 2.6 x 2.7 cm	No significant enlarged cervical LN

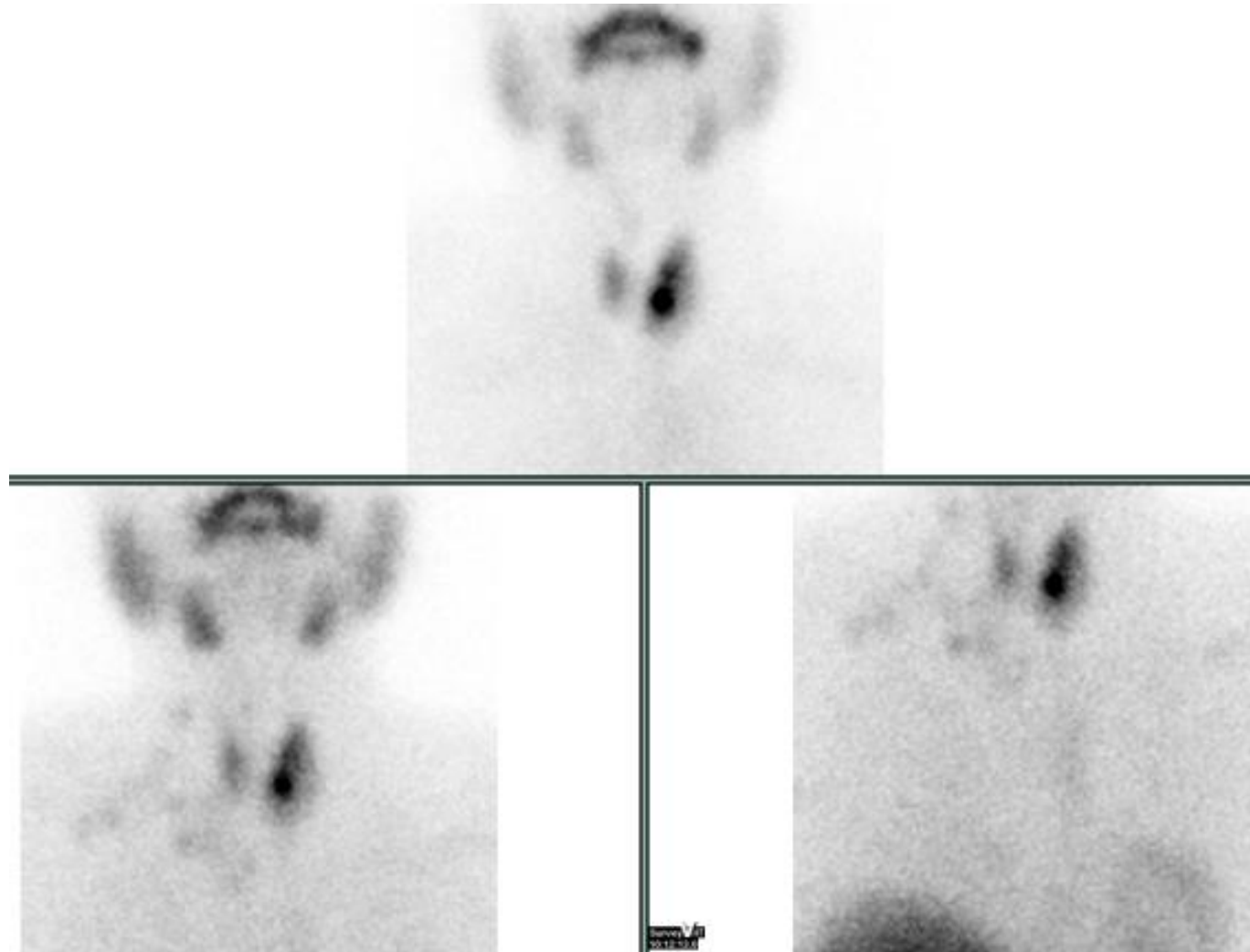
Parathyroid scan

	Rt upper	Rt lower	Lt upper	Lt lower	Note
Parathyroid scan 31/10/65	Less degree of uptake Due to prior thyroidectomy			Non-uniform uptake 3 cm. mixed solid-cystic mass Possibly functioning thyroid nodule/mass	-Irregular areas of increase uptake at rt sided neck, upper mediastinum and rt axillary regions → MIBI- avid LN No definite scan finding of parathyroid adenoma***
Total body scan 4/11/65					-Irregular areas of increase uptake at rt sided neck, upper mediastinum and rt axillary regions → MIBI- avid LN

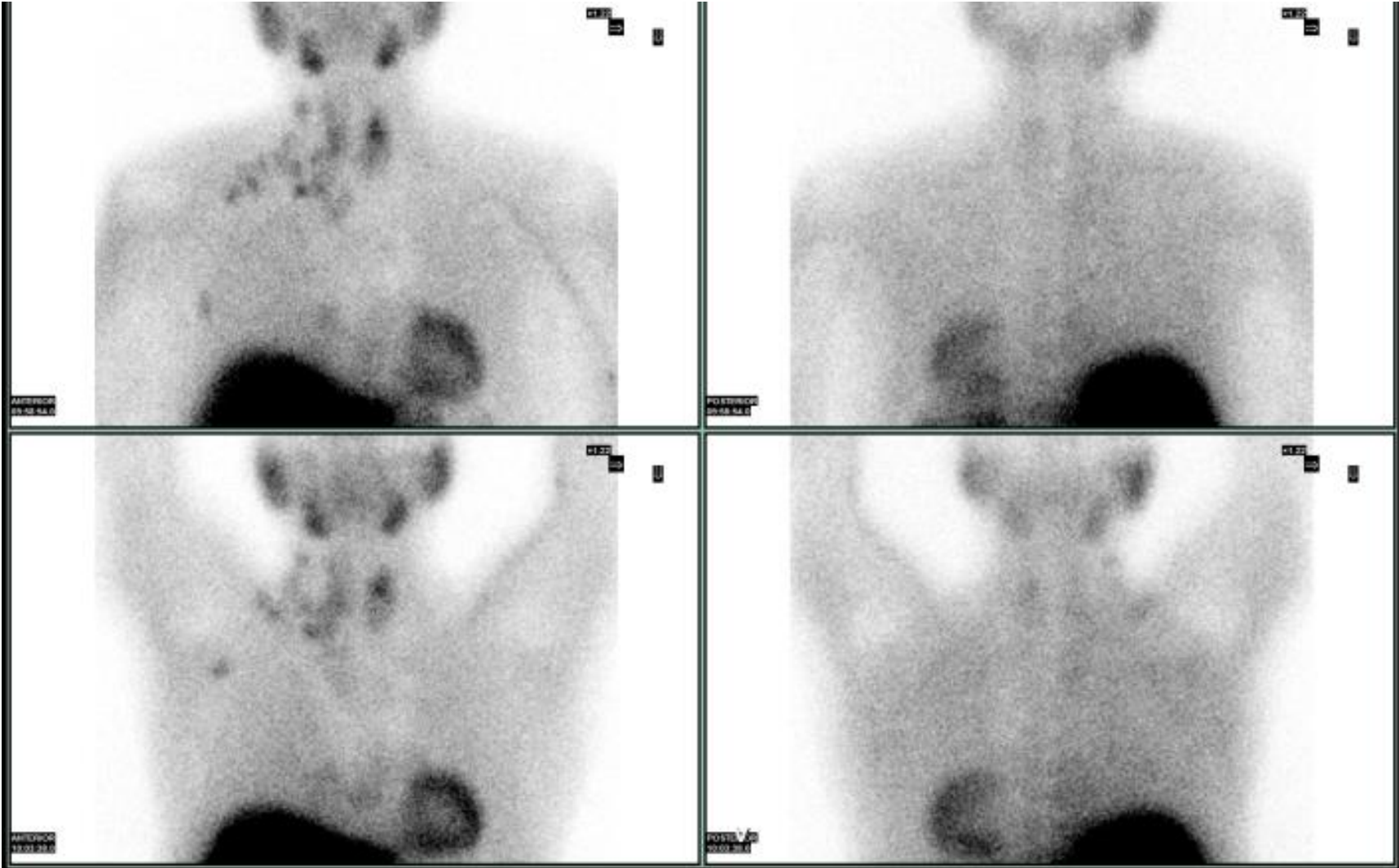
เรียนเชิญ รศ.พญ.ชนิศา โชติพานิช

- ช่วยอ่านผล MIBI scan และ Total body scan
- ช่วยอภิปรายว่าผลการตรวจเข้ากับ parathyromatosis ได้หรือไม่

Parathyroid scan



Total body scan



Genetic testing

- Hyperparathyroidism gene panels (AP2S1, CASR, CDC73, CDKN1B, GNA11, MEN1, RET)
- Result : **no reportable genetic variants**

S/P Left thyroidectomy and Parathyroidectomy 20/12/65

	Rt upper	Rt lower	Lt upper	Lt lower	Note
Operation 20/12/65	<ul style="list-style-type: none"> - ไม่เห็น rt parathyroid glands*** - Remove rt lateral tissue suspected remaining parathyroid glands (nodules) 		Removed nodule 1.3 cm	Removed nodule 1.6 cm	
Intraop frozen	ไม่ได้ส่ง		Parathyroid	Thyroid	
Patho Gross section	- Parathyroid hyperplasia		-Thyroid nodule -No parathyroid tissue		

S/P partial thyroidectomy of left lobe with parathyroidectomy 20/12/65

2nd Admission : Partial thyroidectomy left lobe with parathyroidectomy 20/12/65

Date	Time	Ca	Alb	cCa	iCa	PO4	PTH	Vit D	Mg	Cr/GFR	Mx
15/12/65		13	4.12	12.9		2.2			1.7	1.86	- 2 nd Zolendronate 4 mg iv drip in 30 min
19/12/65		11.3	3.43	11.8		1.9					
20/12/65		11.0	3.38	11.3		2.0			1.1		S/P partial thyroidectomy left lobe with parathyroidectomy 20/12/65 Intra op PTH : 1996>>1987
		11.4		11.4							
21/12/65		11.2	3.3	11.7		2.8	1795				
22/12/65		11.4	3.29	11.9		2.1					
23/12/65		11.6	3.20	12.2		1.8			2.1		-3 rd Zolendronate 4 mg iv drip in 30 min
25/12/65		11.6	3.32	12.1		2.1			1.3		
27/12/65		12.1	3.65	12.3		2.3			2.0		- D/C

3rd Admission : Moderate hypercalcemia, AKI

Date	Time	Ca	Alb	cCa	iCa	PO4	PTH	Vit D	Mg	Cr/GFR	Mx
5/1/66		13.1	3.54	13.46	7.2	3.3	1960	28.9		3.85/13	- Calcitonin 200 IU sc q 6 hrx 2 day -IV NSS
6/1/66		13.3	3.21	13.9						2.93/14	
7/1/66		12.6	3.32	13.1						2.28/25	-4 th Zolendronate 4 mg iv drip in 30 min

Question 4: ท่านจะวางแผนการรักษาผู้ป่วยรายนี้อย่างไรต่อไป (F2)

Management Plan

- Refractory hypercalcemia
- Persistent PHPT

3rd Admission : Moderate hypercalcemia, AKI

Date	Time	Ca	Alb	cCa	iCa	PO4	PTH	Vit D	Mg	Cr/GFR	Mx
5/1/66		13.1	3.54	13.46	7.2	3.3	1960	28.9		3.85/13	- Calcitonin 200 IU sc q 6 hrx 2 day -IV NSS
6/1/66		13.3	3.21	13.9						2.93/14	
7/1/66		12.6	3.32	13.1						2.28/25	-4 th Zolendronate 4 mg iv drip in 30 min
11/1/66		11.4	3	12.2		2.4			1.6	1.9/30	- start Cinacalcet(25) 1x2 po pc
14/1/66		11.8	3.24	12.4		2.5				2.0/28	-5 th Zolendronate 4 mg iv drip in 30 min
16/1/66		10.2	3.28	10.77		2.3					
18/1/66		10.2	3.45	10.6		2.8				2.4/23	-increase Cinacalcet(25) 1x3 po pc
20/1/66		9.8	3.3	10.3		2.3					
22/1/66		9.7	2.99	10.5		2.5				2.24/25	
3/2/66		9.3	2.89	10.1							-D/C with Cinacalcet(25) 1x3 po pc

Re-evaluate 4DCT

	Rt upper	Rt lower	Lt upper	Lt lower	Note
4dCT	unremarkable	No significant change of a well-defined hypodense nodule 0.5x1.0x0.8 cm Favor parathyroid adenoma	Disappear of previously hypodense nodule at posterior aspect No significant change of well-defined hypodense nodule 0.4x0.7x0.8 cm Favor LN	Evidence of lobectomy at left thyroid lobe and isthmus with few residual thyroid tissue at left thyroid bed	

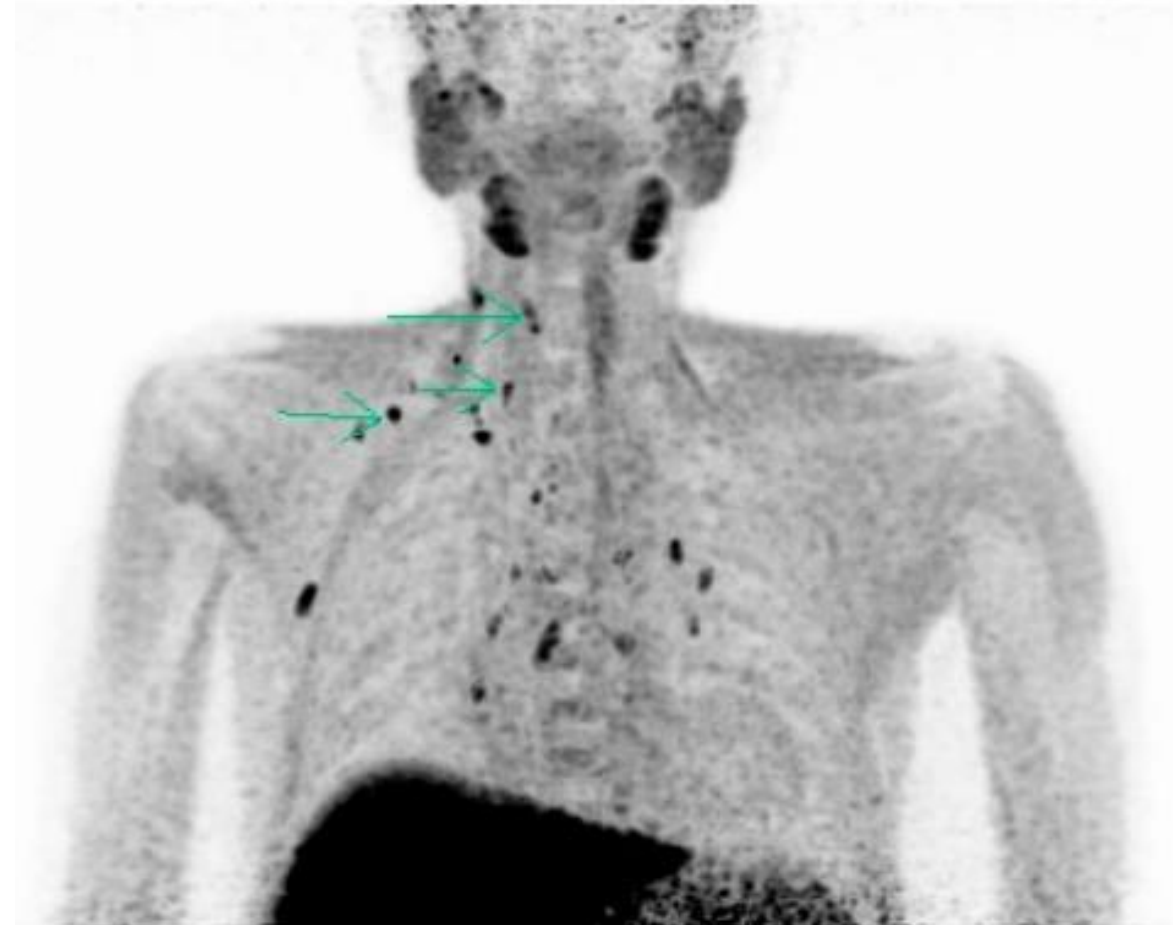
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- ช่วย Discuss การส่งตรวจ MIBI scan และ PET/CT ใน Primary hyperparathyroidism และ Parathyromatosis
- ช่วยอ่านผล PET/CT

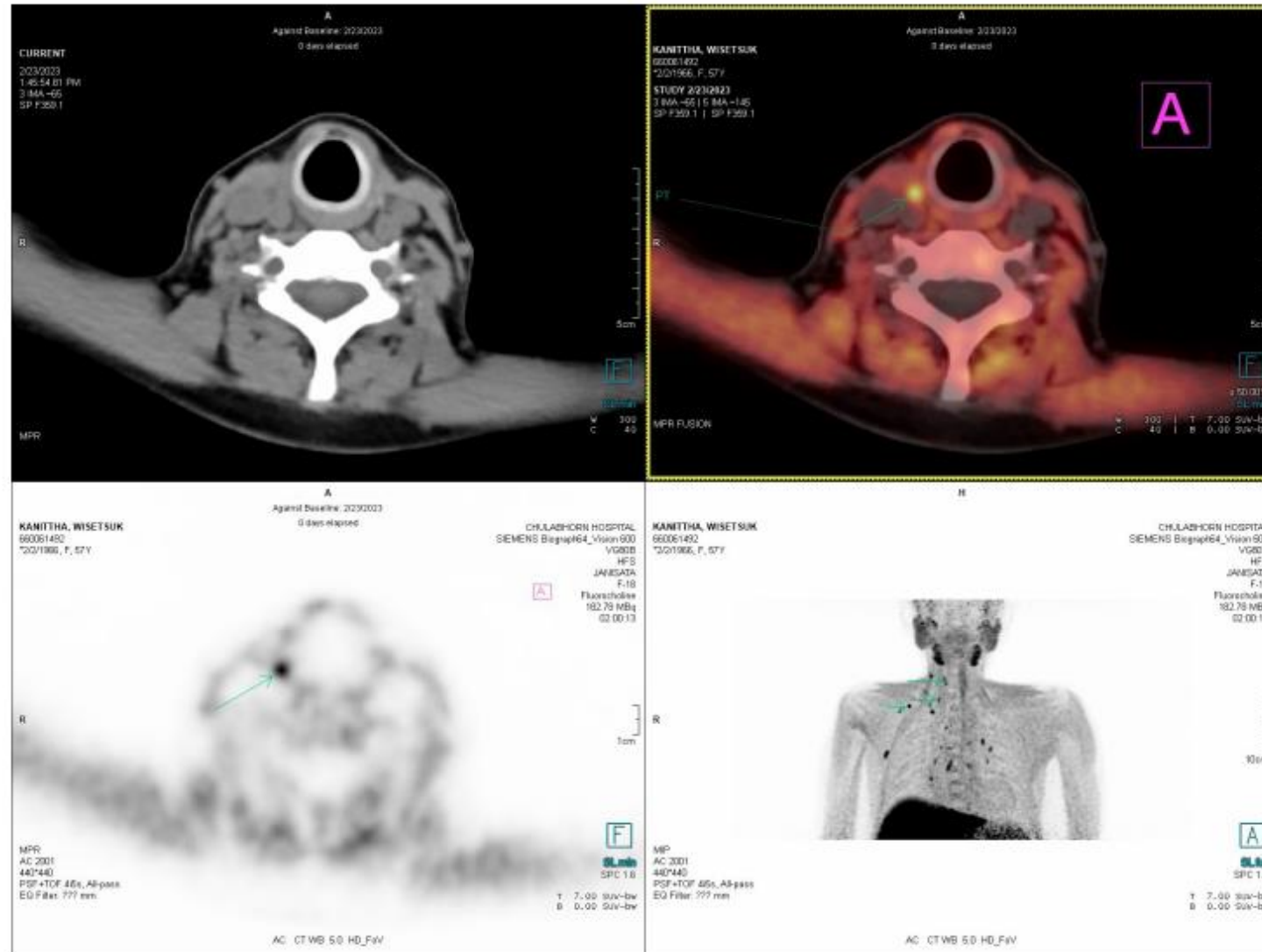
PET CT

18F-FLUOROCHOLINE PET/CT INTERPRETATION:

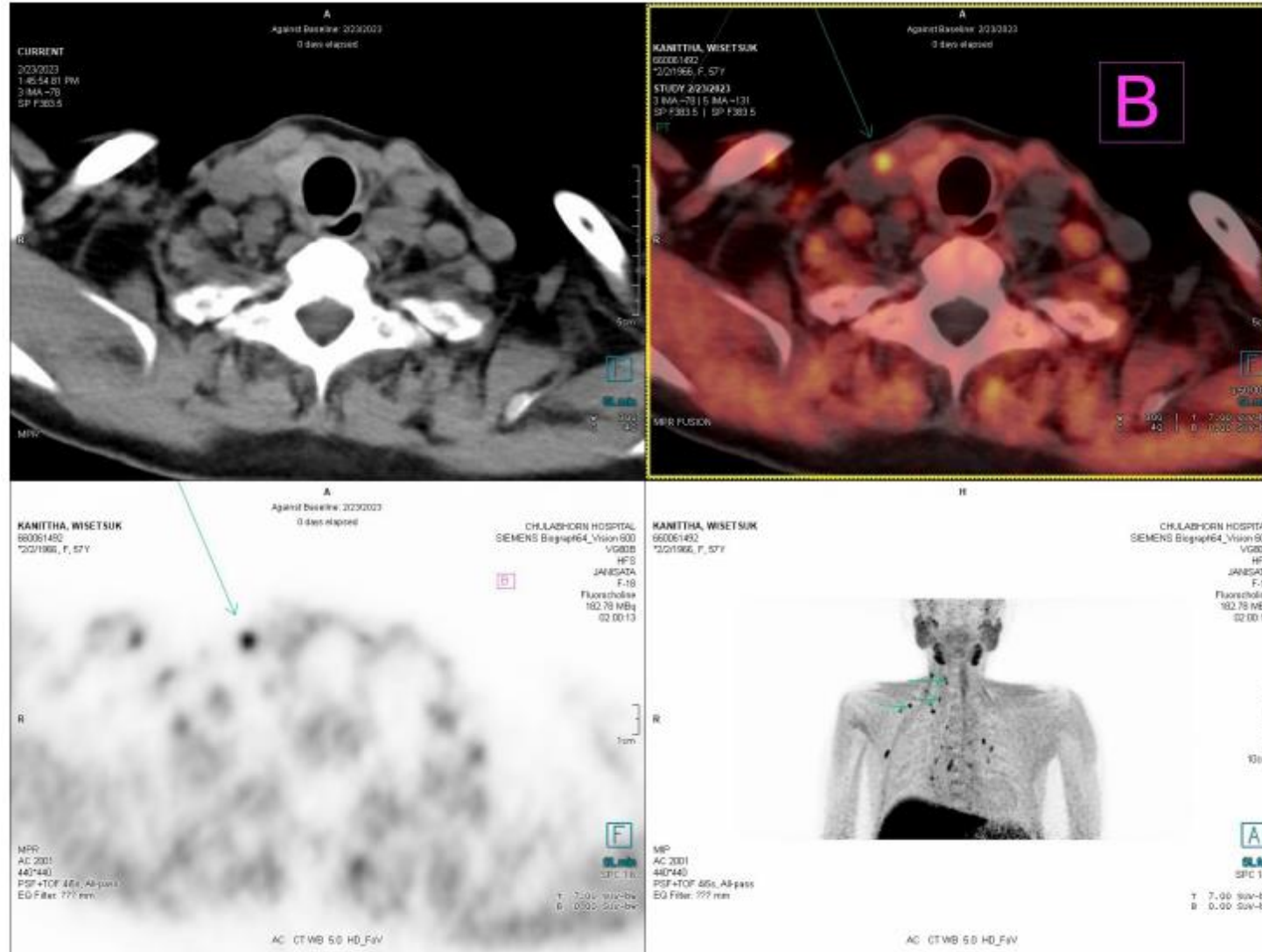
- A 0.6-cm nodule with fluorocholeline uptake at posterior aspect of **upper pole of the right thyroid lobe**; **hyperfunctioning parathyroid gland is likely**
- A 0.6-cm nodules with fluorocholeline uptake **at lateral and posterior aspect of lower pole of the right thyroid lobe**; **hyperfunctioning parathyroid glands are possible**
- Multiple nodule-like lesions with fluorocholeline uptake at **right cervical (level III-IV), right supraclavicular, right infraclavicular, mediastinal and right axillary regions**, up to 0.7 cm in short axis; **pathologic nodes are possible**.
DDx reactive nodes
- A 0.5-cm skin nodule with mild fluorocholeline uptake



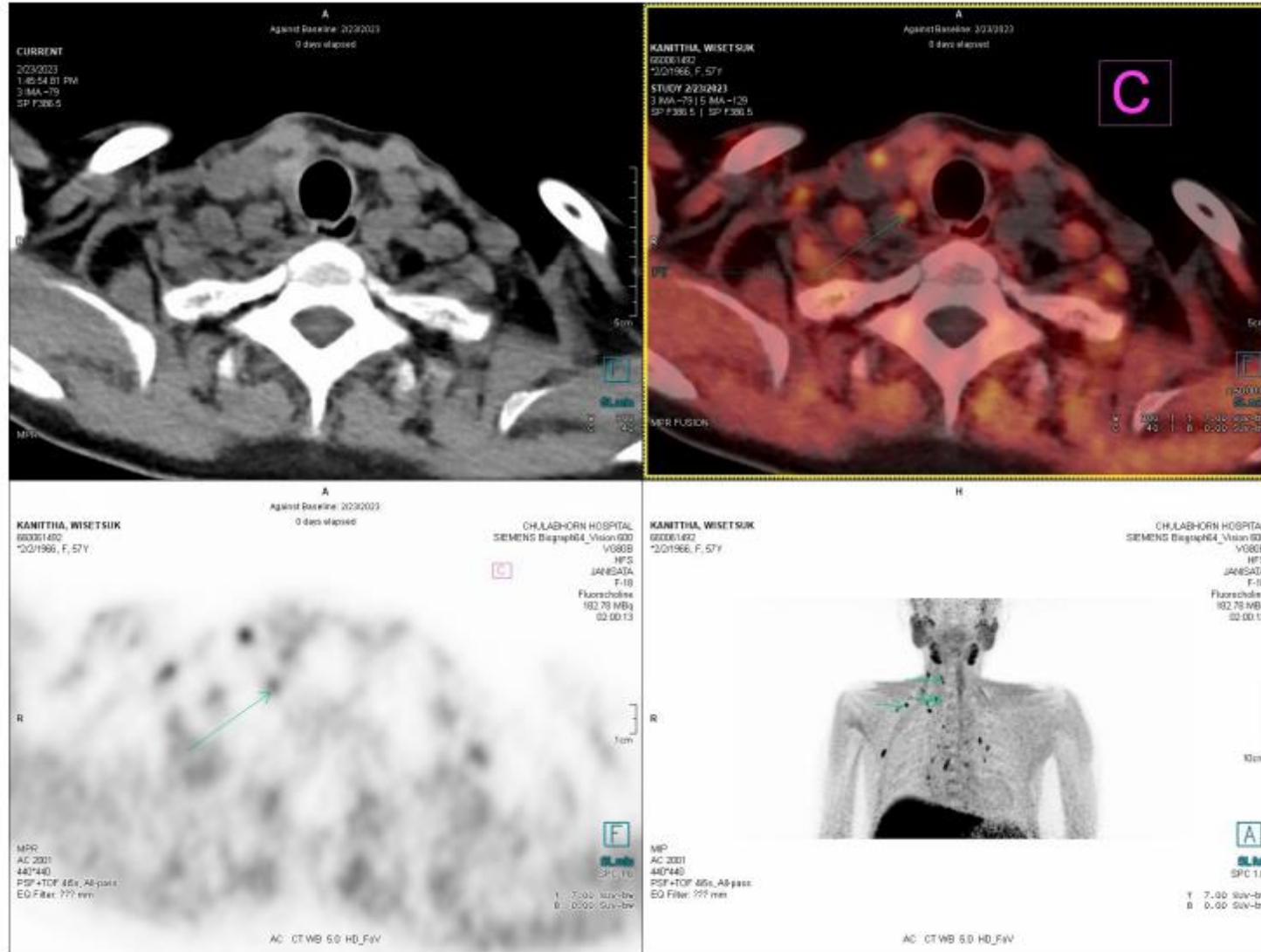
PET CT



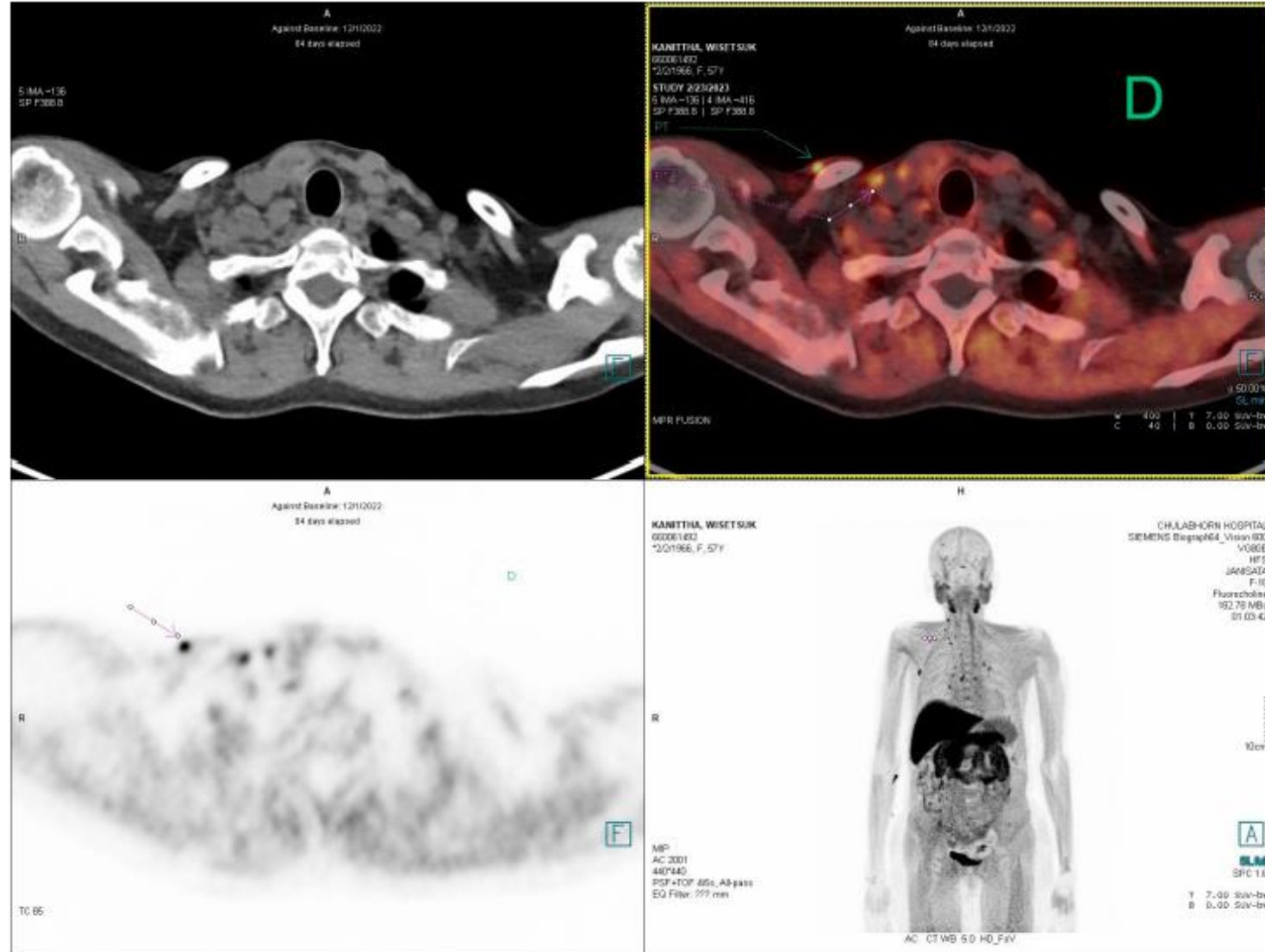
PET CT



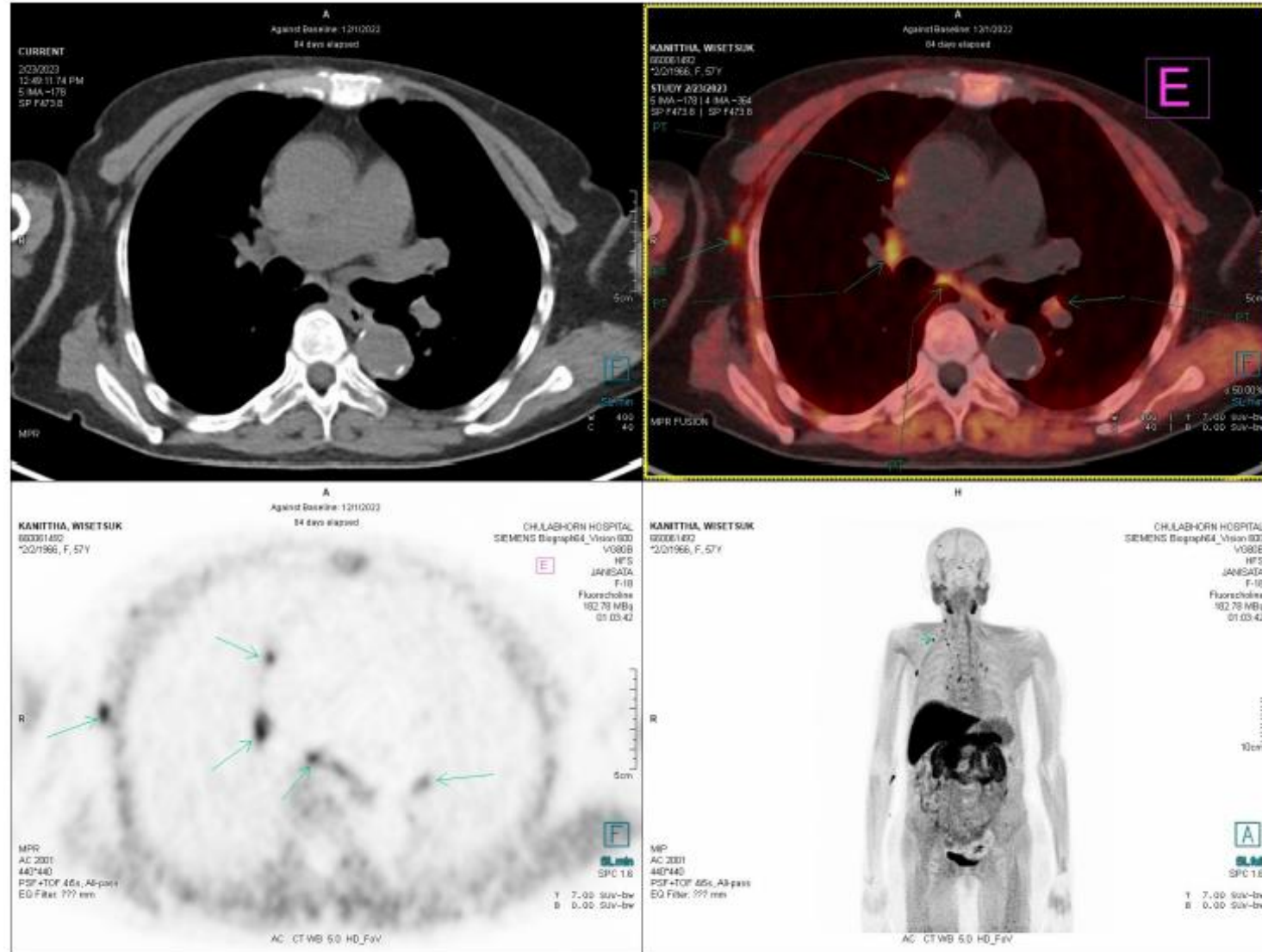
PET CT



PET CT



PET CT



S/P right parathyroidectomy with soft tissue excision with intraoperative frozen section 7/3/66

	Rt upper	Rt lower	Lt upper	Lt lower	Other
Operation 7/3/66	No parathyroid	Removed nodule	ไม่เห็น left parathyroid glands***		-Removed soft tissues of Rt chest parathyroid, Rt axillary
Intraop frozen	2 Lymph node	1 parathyroid tissue 2 Lymph node	ไม่ได้ส่ง		- Chest : confirm 1 parathyroid tissue -Rt axillary : ไม่ได้ส่ง
Patho Gross section		Parathyroid tissue (0.1-0.4 cm)			Chest : focal fibrosis Rt axillary dissection: - Lymph node 7(negative for malignancy) - Parathyroid tissue1 IHC : Galectin-3 negative, Ki67 2-3% : favor parathyroid adenoma>carcinoma

S/P right parathyroidectomy with soft tissue excision with intraoperative frozen section 7/3/66

Date	Time	Ca	Alb	cCa	iCa	PO4	PTH	Vit D	Mg	Cr/GFR	Mx
1/3/66		9.1	2.67	10.1							-Cinacalcet(25) 1x3 po pc
5/3/66		9.8	3.14	10.4							
7/3/66		9.2	3.02	9.98	2.2					1.65/36	S/P right parathyroidectomy with soft tissue excision with intraoperative frozen section 7/3/66 Intra op PTH : 840>>937 -off cinacalcet
8/3/66		9.5	2.96	10.3			743				
11/3/66		10.6	2.95	11.4			931			1.41/44	-6 th Zolendronate 4 mg iv drip in 30 min
14/3/66		11.2	3.12	11.9							
17/3/66		12.1	3.06	12.8							-7 th Zolendronate 4 mg iv drip in 30 min
19/3/66		11	3.34	11.5							
20/3/66		12.1	3.5	12.5							- restart Cinacalcet(25) 1x2 po pc
22/3/66		11.4	3.28	11.9							
24/3/66		11.5	3.4	11.9						1.53/39	-D/C with Cinacalcet(25) 1x2 po pc

Progression

- **Persistent PHPT from parathyromatosis**
 - Control resistant hypercalcemia → cinacalcet (25) 1x2 po pc (calcium level 11-11.8 mg/dL)
 - 4dCT and parathyroid scan +/- re-operation
- Hypothyroidism
 - L-thyroxine 400 mcg/week

Parathyromatosis

14th July 2023

Parathyromatosis

- A rare cause of recurrent PHPT
- The presence of nodules of hyperfunctioning parathyroid tissue outside the anatomical confines of the parathyroid glands
- Usually in the neck or mediastinum

Parathyromatosis

- **Type 1** : Hyperplasia of embryologic remnants of parathyroid tissue
- **Type 2** : Spillage and seeding of parathyroid cells as a complication of surgical interventions involving the parathyroids or the thyroid gland
 - Duration between first surgery : reported 3-29 year
 - Ca level : reported 11-15.6 mg/dL
 - PTH level : reported 107- >3480 pg/ml , 7-23 times higher than normal values

Distinguishing Parathyromatosis, Atypical Parathyroid Adenomas, and Parathyroid Carcinomas Utilizing Histologic and Clinical Features

Jefree J. Schulte^{1,2} · Garrison Pease³ · Jerome B. Taxy⁴ · Curtis Hall⁴ · Nicole A. Cipriani¹

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- N= 27
- No difference in serum PTH levels(P=0.90) :
 - Parathyromatosis (615 pg/mL, range 266–1992 pg/mL)
 - Atypical adenoma (601 pg/mL, range 101–2518 pg/mL)
 - Parathyroid carcinoma (957 pg/mL, range 79–2250 p/mL)
- No difference in serum calcium levels(P=0.40) :
 - Parathyromatosis (13.2 mg/dL, range 12.8–13.6 mg/dL)
 - Atypical adenoma (11.5 mg/dL, 9.3–15.6 mg/dL)
 - Parathyroid carcinoma (11.8 mg/dL, range 11–14.2 mg/dL)

Histopathology

- **Parathyroid carcinoma** : invasive growth, either invasion into surrounding tissues, blood vessels, nerves, or by the presence of distant metastatic disease
- **Parathyromatosis and Atypical adenoma** : no manifest distant metastases

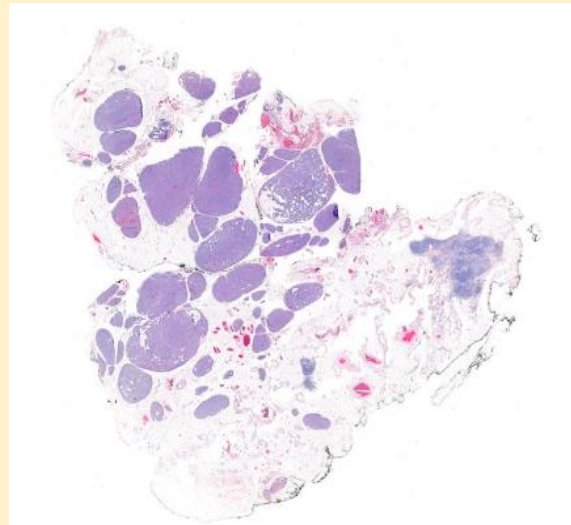


Fig.5 Case of parathyromatosis showing "buckshot" scattering of numerous well-circumscribed nodules of benign parathyroid tissue (H&E, 20X)

Nested soft tissue invasion

- **FNA :**

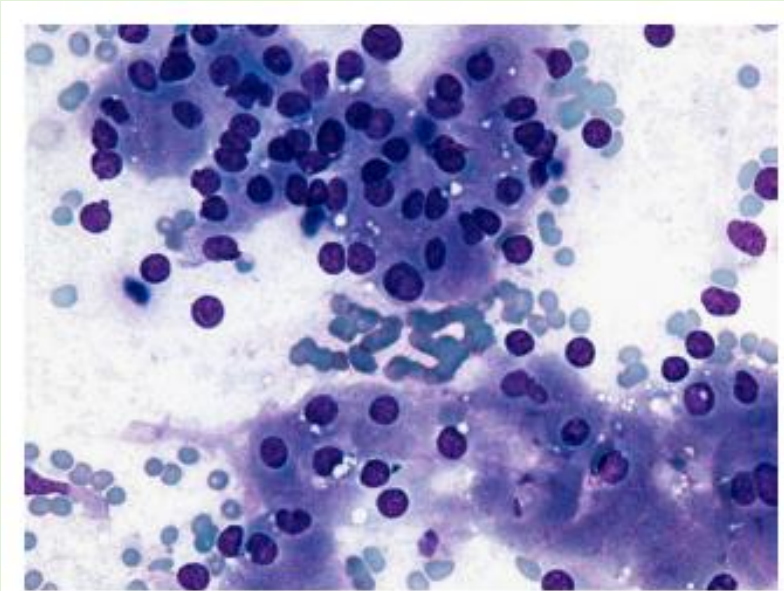


Fig. 12 – Parathyroid cytology: FNA of parathyroid lesion showing a monotonous population of oncocytic cells arranged in trabeculae (Diff-Quik[®] stain). On the basis of morphology, this FNA specimen cannot be distinguished from an oncocytic thyroid lesion. The PTH levels performed on this aspirate were high and confirmed the diagnosis.

Histopathology

- Immunohistochemistry

- The studies showed diffuse reactivity for PTH, GATA3, and chromogranin A,
- Non-reactivity for TTF1, GFAP, and CK7
- **KI67 >5%** ; Favor carcinoma

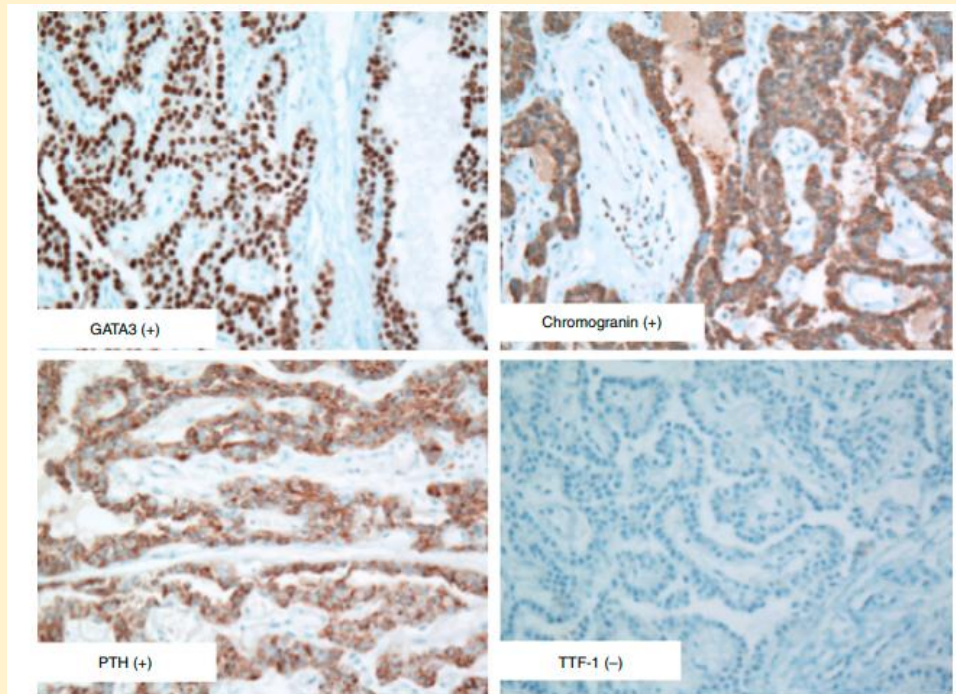
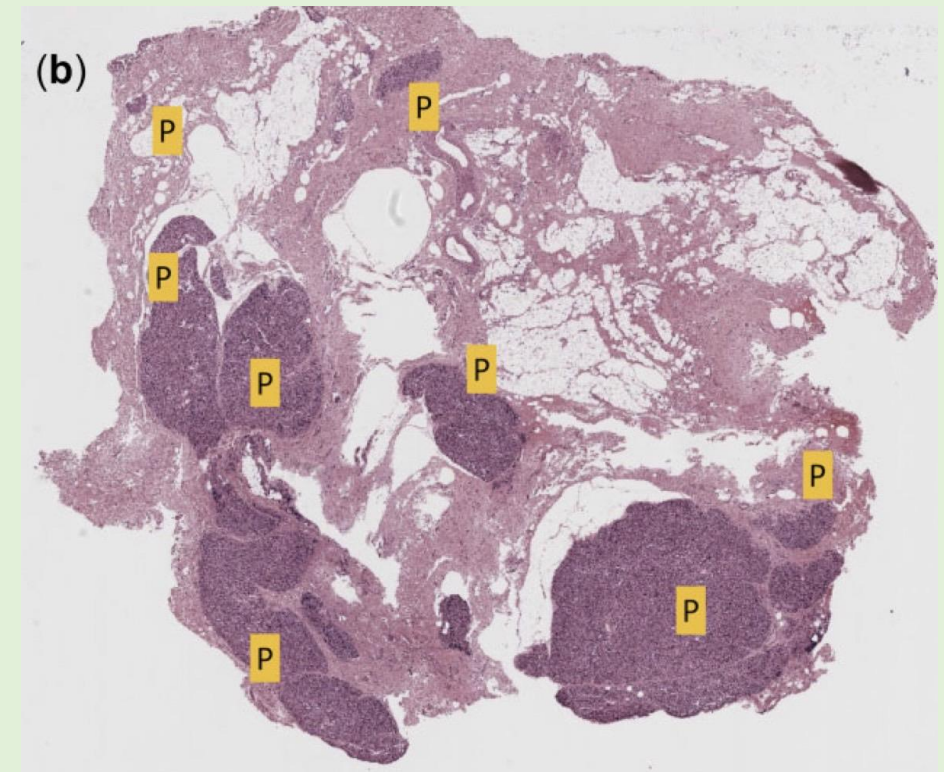


Figure 5. Immunohistochemistry. Immunohistochemical stains for GATA-3 (upper left), chromogranin (upper right) and parathyroid hormone (PTH) (lower left) are all positive, and thyroid transcription factor-1 (TTF-1) staining (lower right) is negative, supporting a parathyroid origin.

- Pathology :



multiple and juxtaposed islets of parathyroid tissue

Imaging

- **The first-line imaging modalities : Ultrasound and parathyroid scintigraphy**
 - Sensitivity of 73–90% and 78–90%
 - Positive predictive value (PPV) of 86–93% and 83–94%
- The concordance of both imaging modalities sufficient for guiding reoperation
- **4DCT** : use in cases of negative, discordant or indeterminate results on the first-line imaging studies

- **Functional and anatomical imaging**
 - **11C-methionine PET/CT**
 - **18F-choline PET/CT**
- **If all non-invasive procedures are negative**
 - **Selective venous sampling** with assessment of PTH can correctly localize the anatomical region ,sensitivity 73 to 100%

Management

- **Medical treatment**

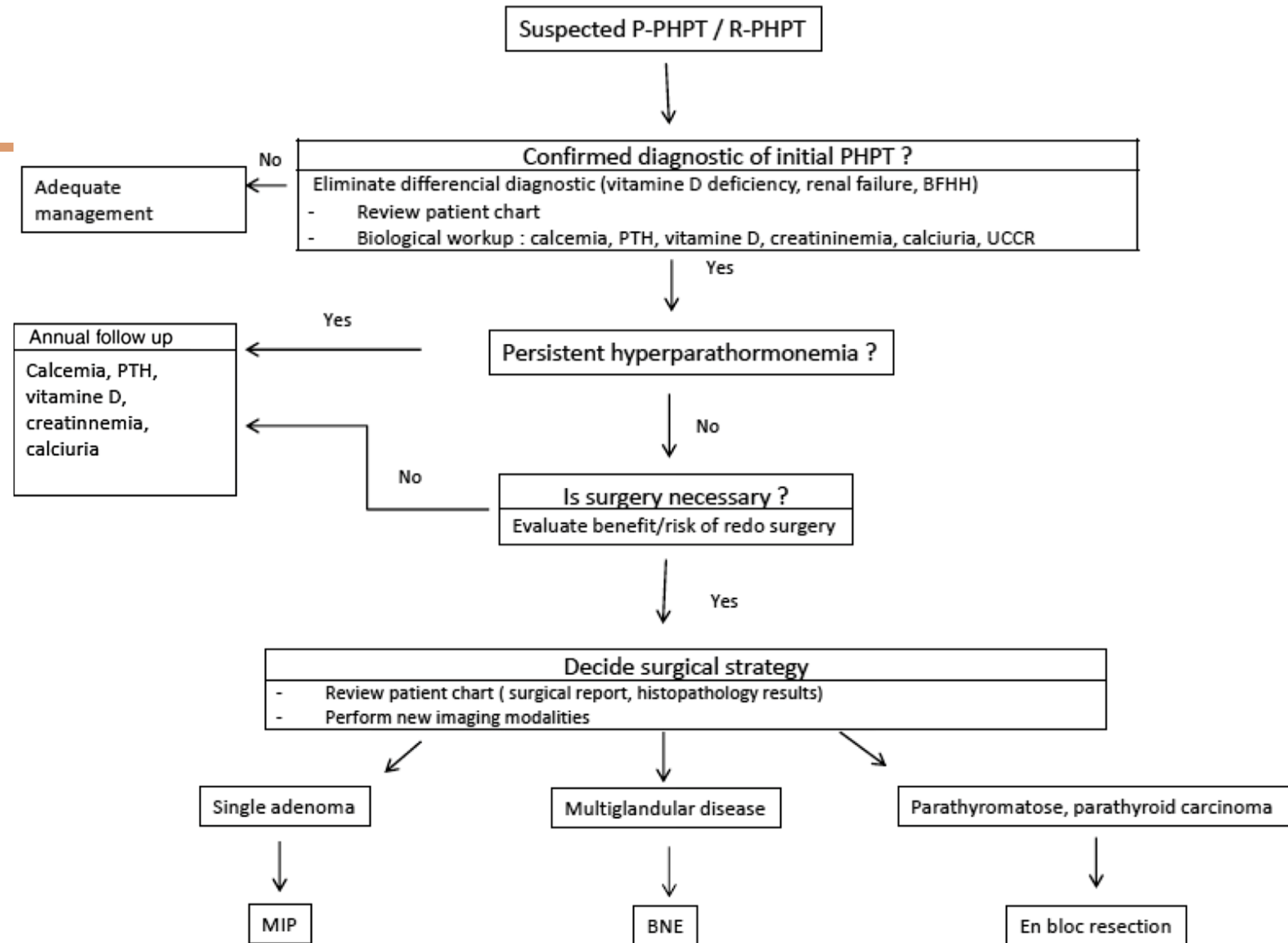
- Cinacalcet
- Antiresorptive :
Bisphosphonate,
Denosumab

- **Surgery**

- En-bloc surgery

- **Other :**

- Microwave ablation
- Percutaneous ethanol ablation



Management

Table 1 Reported parathyromatosis cases in patients with primary hyperparathyroidism from 1975 to September 2018 in English literature.

Authors, Year (Reference number)	Cases (n)	Treatment	Outcome
Reddick <i>et al</i> , 1977 ³	1	3 reoperations	Remission
Rattner <i>et al</i> , 1985 ⁴	2	1 and 3 reoperations	Remission
Akerstrom <i>et al</i> , 1988 ⁵	3	1/2/2 reoperations	Remission
Sokol <i>et al</i> , 1993 ⁶	1	2 reoperations	Persistence
Evans <i>et al</i> , 2005 ⁷	1	1 reoperation	Remission
Tublin <i>et al</i> , 2007 ⁸	1	2 reoperations	Remission
Diaconescu <i>et al</i> , 2011 ⁹	1	1 reoperation	Remission
Mohammadi <i>et al</i> , 2012 ¹⁰	1	1 reoperation	Remission
Hage <i>et al</i> , 2012 ¹¹	1	4 reoperations + cinacalcet	Mild remission
Twigt <i>et al</i> , 2013 ¹²	2	1 and 3 reoperations	Remission
Pinnamaneni <i>et al</i> , 2013 ¹³	1	2 reoperations	Persistence
Scorza <i>et al</i> , 2014 ¹⁴	1	3 reoperations + cinacalcet	Persistence
Edling <i>et al</i> , 2014 ¹⁵	1	1 reoperation + cinacalcet	Remission
Sharma <i>et al</i> , 2016 ¹⁶	1	1 reoperation	Persistence
Jain <i>et al</i> , 2017 ¹⁷	1	2 reoperations	Remission
Aggarwal <i>et al</i> , 2017 ¹⁸	1	1 reoperation	Remission

Parathyromatosis

Case report :

- A 48-year-old man refer to Panagia hospital , Greece
- Presented with a palpable left-sided cervical-> diagnosed with **primary hyperparathyroidism** at the age of 33 (in 2003)
- Serum calcium 19 mg/dl (8.4–10.5), PTH levels at 88 pg/ml (15–65), present bilateral nephrolithiasis
- **Left thyroid lobectomy and removal left lower parathyroid gland by a general surgeon**
- Excision was reportedly technically difficult due to the presence of adhesions to surrounding tissues
- **Histological examination :**
 - 3.5-cm left parathyroid adenoma in addition to six further fragments 1.2–2.6 cm
 - No vascular infiltration or other features suggestive of malignancy were evident

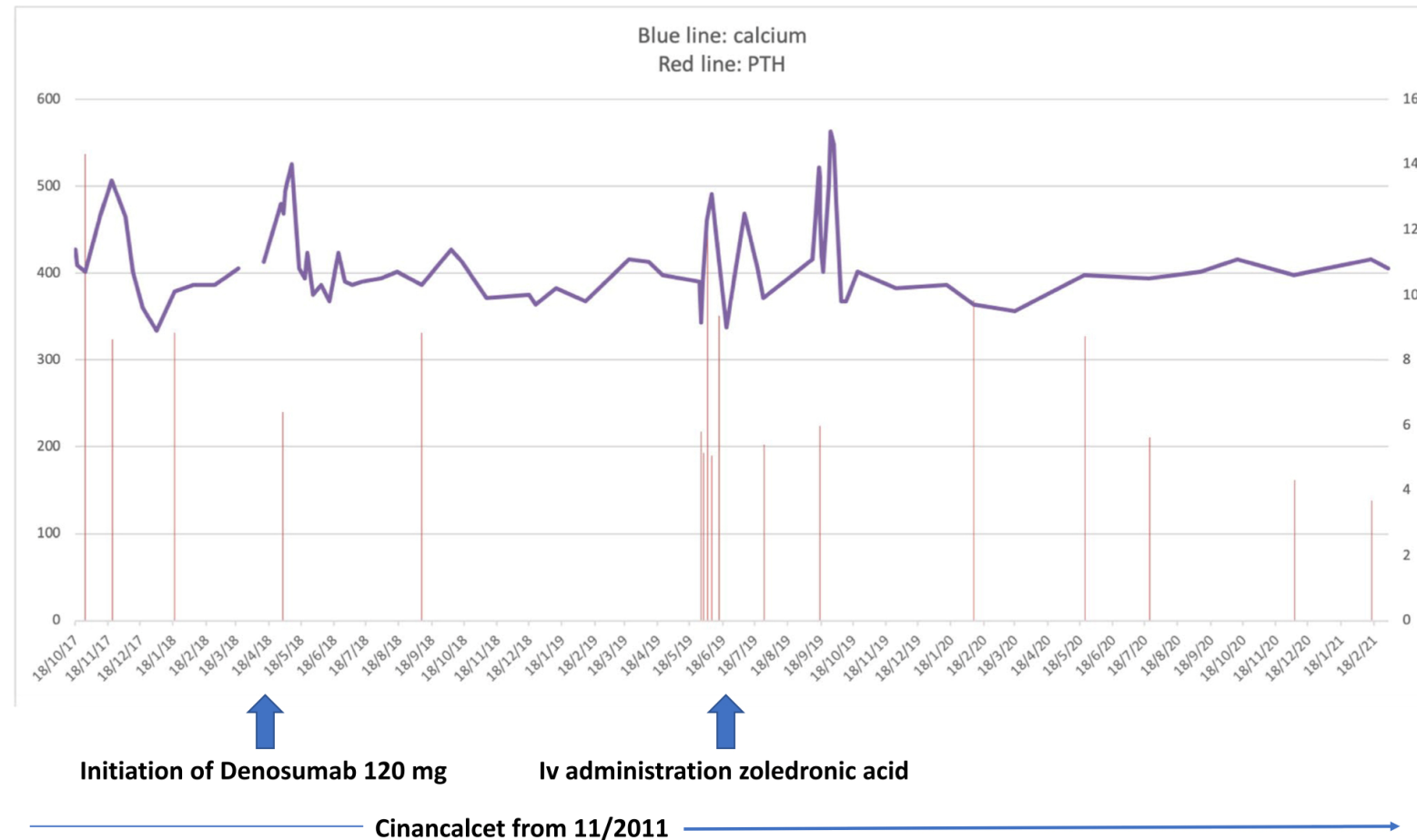
Parathyromatosis

- **Relapse of PHPT Six years** after the initial parathyroidectomy
- Genetic testing for MEN1 and MEN 2A was negative
- **Repeat neck exploration in 2009** , the patient remained normocalcemic for 12 months

- **In February 2011**, recurrent started **cinacalcet** at a dose of 60 mg tid
 - serum calcium and PTH remained elevated (calcium 12.04 mg/dl, PTH 130 pg/ml)
 - surgical excised en bloc, repeated bilateral neck explorations (2012, 2014, 2015)
 - **Risedronate** 35 mg once weekly was added therapy and **uptitration of the cinacalcet maximum recommended dose of 270 mg/day** (2014 to 2016) (PTH 314.8 pg/ml, calcium concentrations 11.12–11.54 mg/ dl)

Parathyromatosis

- In October 2017, serum calcium at the time was 13.1 mg/dl, PTH 217 pg/ml (12–88)
 - Surgical intervention was not feasible
 - Offered an **off-label use of denosumab** and **cinacalcet was discontinued**
 - Serum calcium concentrations increased to pretreatment levels before the end of the month after the injection (12.71 mg/dl)
- In May 2018, decision was taken to use **denosumab at a dose of 120 mg/month combination with cinacalcet**
 - Serum Ca concentrations decreased within the first 2 weeks
 - The combination over a 36-month period was well tolerated without any side effects
 - Maintaining the patient's serum calcium at 9.51–10.54 mg/dl
 - PTH concentrations remained elevated





คณะวิทยาศาสตร์ มหาวิทยาลัยธรรมศาสตร์



Thank You