

Cutaneous metastases of infantile choriocarcinoma can mimic infantile hemangioma both clinically and radiographically

Case Report

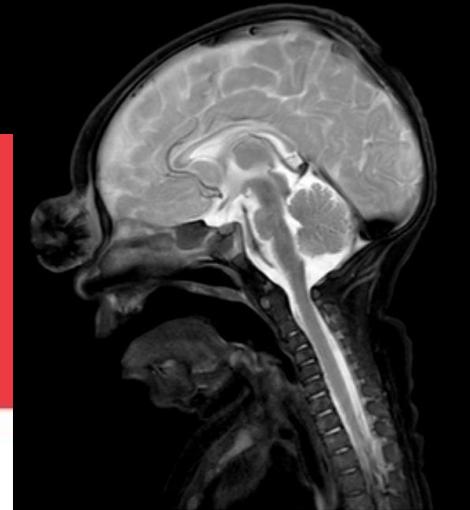
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Disclosures

- None

Objectives

- Understand the clinical and radiographic challenge of distinguishing between a common infantile hemangioma and a much less common solitary cutaneous metastasis
- Understand the time-critical nature of accurate diagnosis of infantile choriocarcinoma
- Understand the unique predilection of infantile malignancies to metastasize to the skin, most commonly lymphoma/leukemia
- Know when to suggest an alternate diagnosis despite the initial clinical presumption of infantile hemangioma

Initial Presentation – 37 day old male

- Initially noticed a flat, blue lesion at 10 days of age (not shown)
- Presents to dermatology clinic with pedunculated, friable red/blue glabellar mass (shown) at 37 days of age
- Otherwise healthy



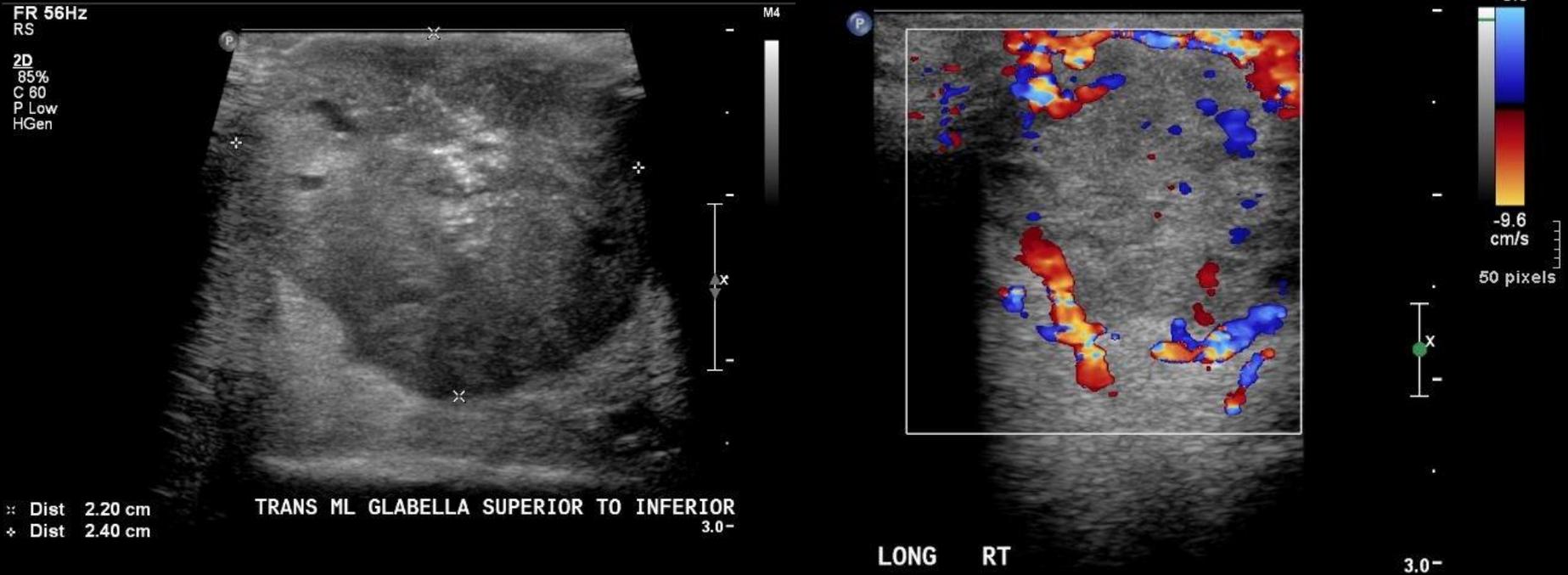
Initial Treatment and Course

- Propranolol prescribed for presumed infantile hemangioma
- Lesion rapidly grew in size over several weeks
- Mother concerned with alarming growth rate and extension into both eyes affecting lid closure
- Patient sent to ER, ultrasound ordered to exclude deeper extension
- MRI also ordered due to history of bronchiolitis and concern for possible airway hemangioma





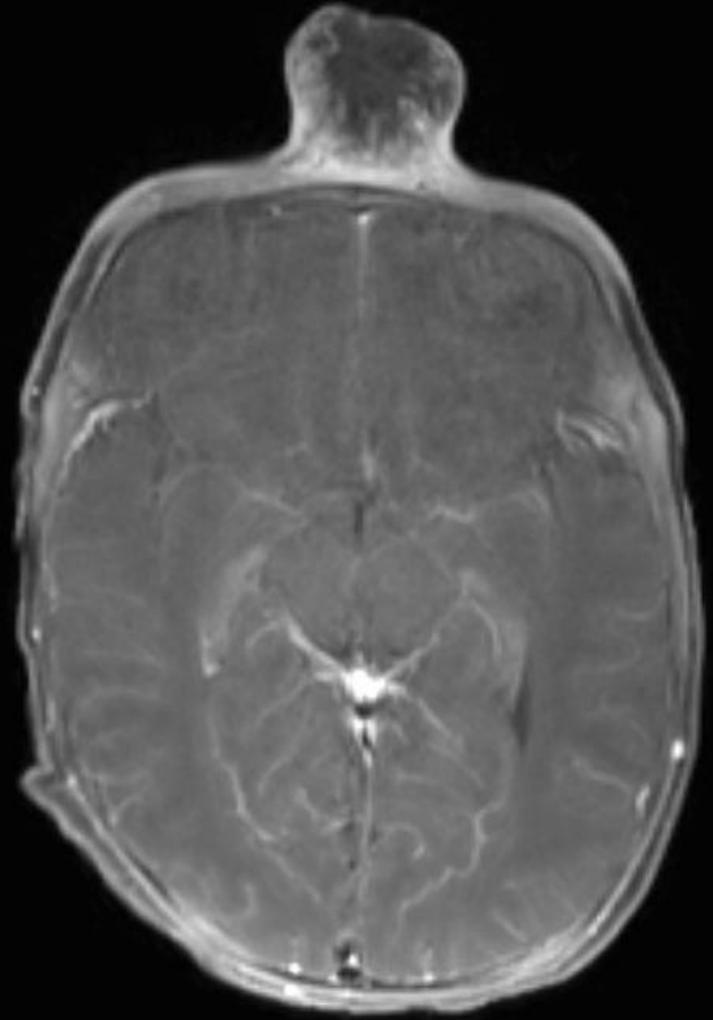
Initial ultrasound



Solid hypervascular 2.4 x 2.2 cm mass interpreted as
“consider infantile hemangioma or tufted angioma.”



Sagittal T2: Heterogeneous,
centrally hypointense



Axial T1 post-contrast:
Peripheral enhancement

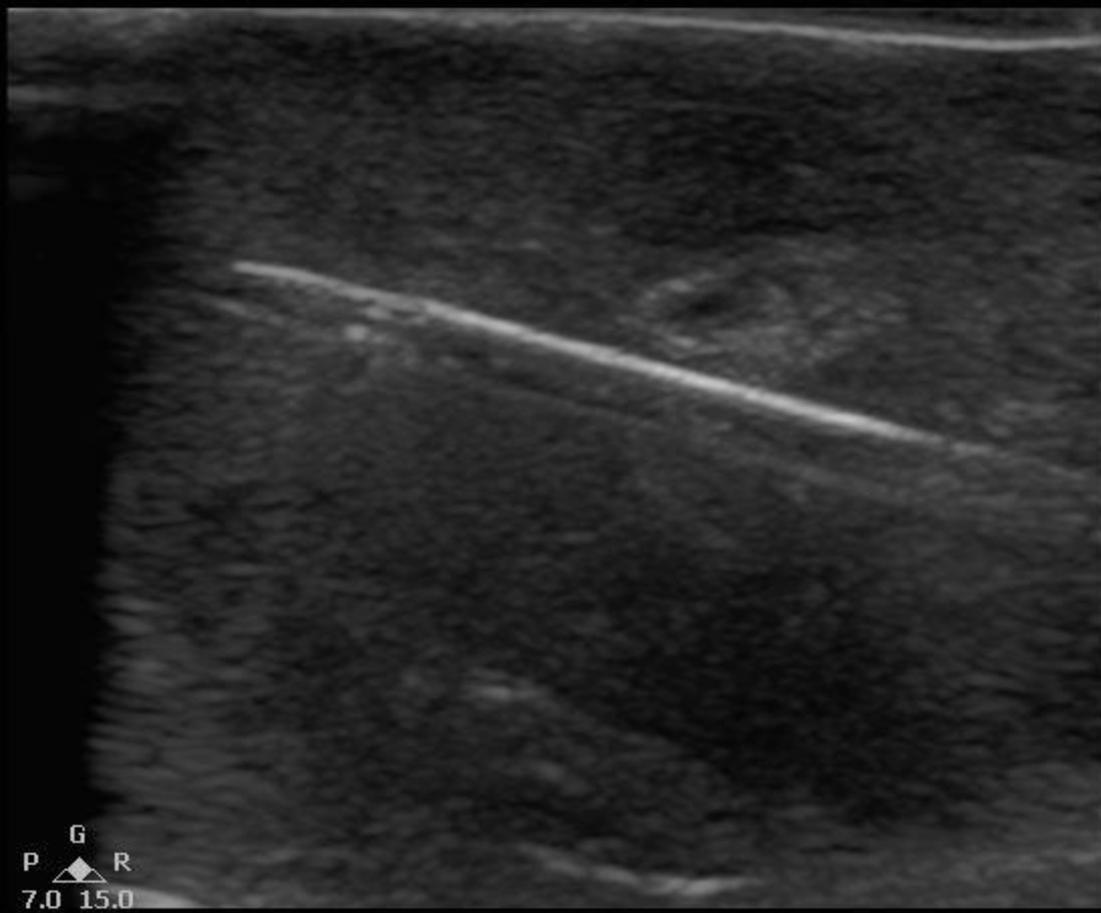
MRI results

- No airway or neck hemangioma(s)
- Although some MRI features not characteristic of clinical diagnosis of infantile hemangioma, the exam was interpreted as “vascular mass arising from glabella. Coordinate with upcoming biopsy to confirm diagnosis of hemangioma.”

Initial work-up

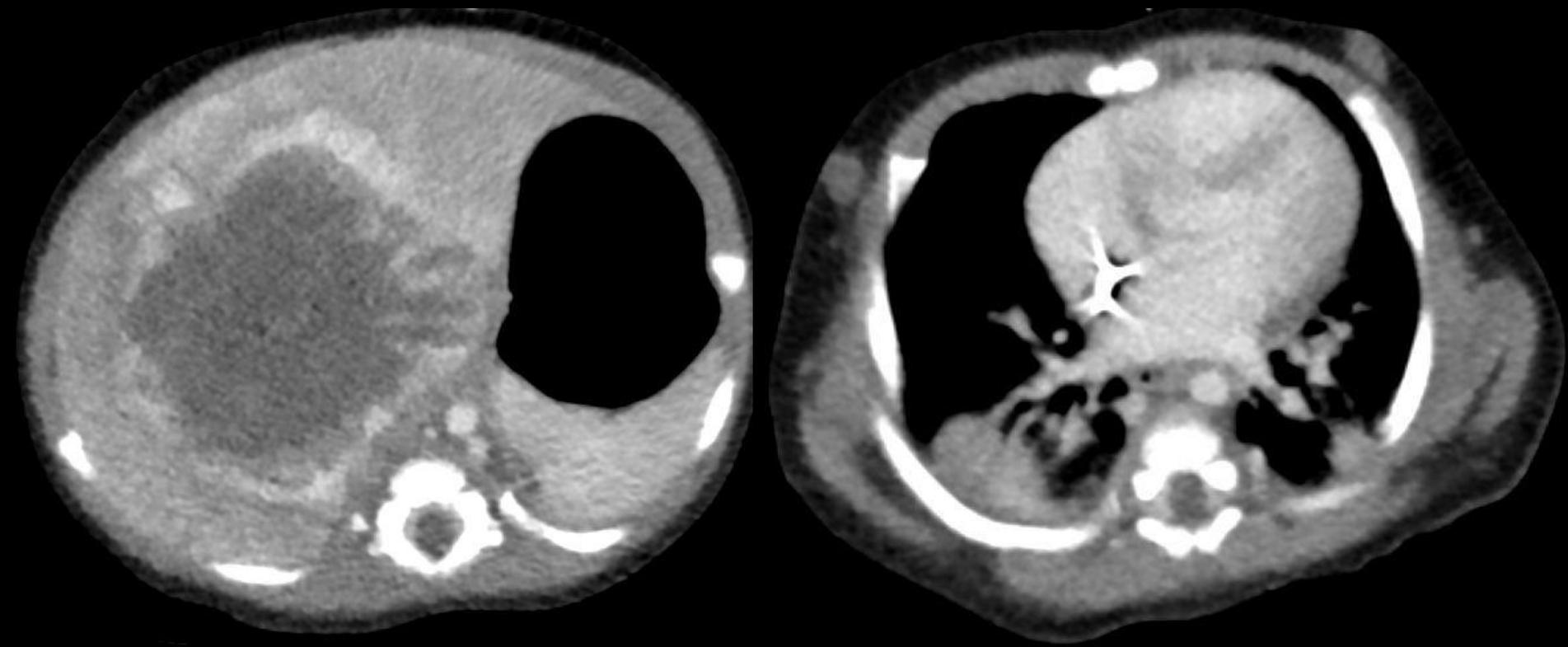
- Lab testing: anemia (7.4 mg/dL) and low platelets (95 K/uL)
- Persistent tachycardia, tachypnea
- Differential at this time:
 - tufted angioma
 - infantile hemangioma
 - kaposiform hemangioendothelioma with concern for possible Kasabach-Merritt syndrome
- Biopsy requested

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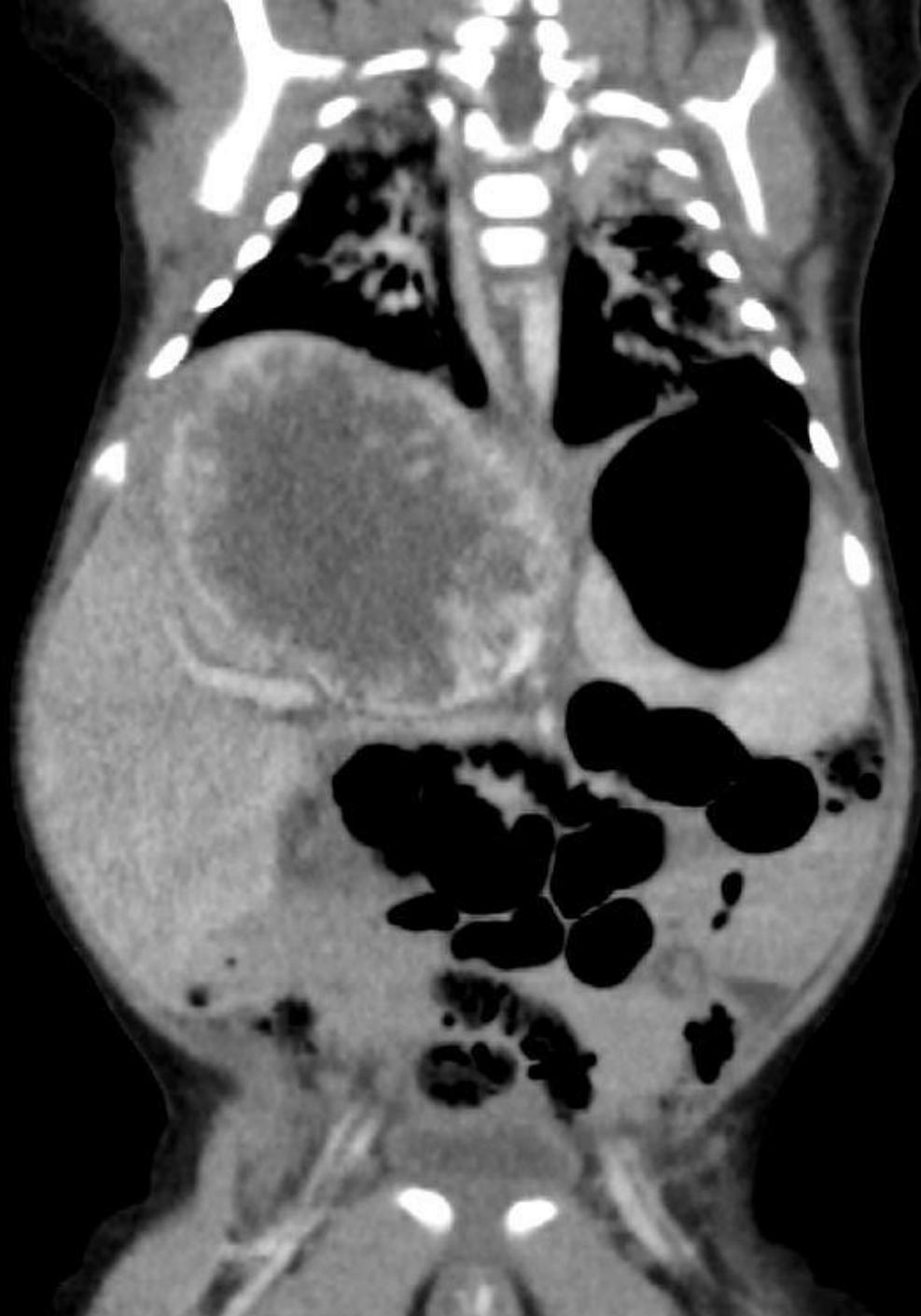


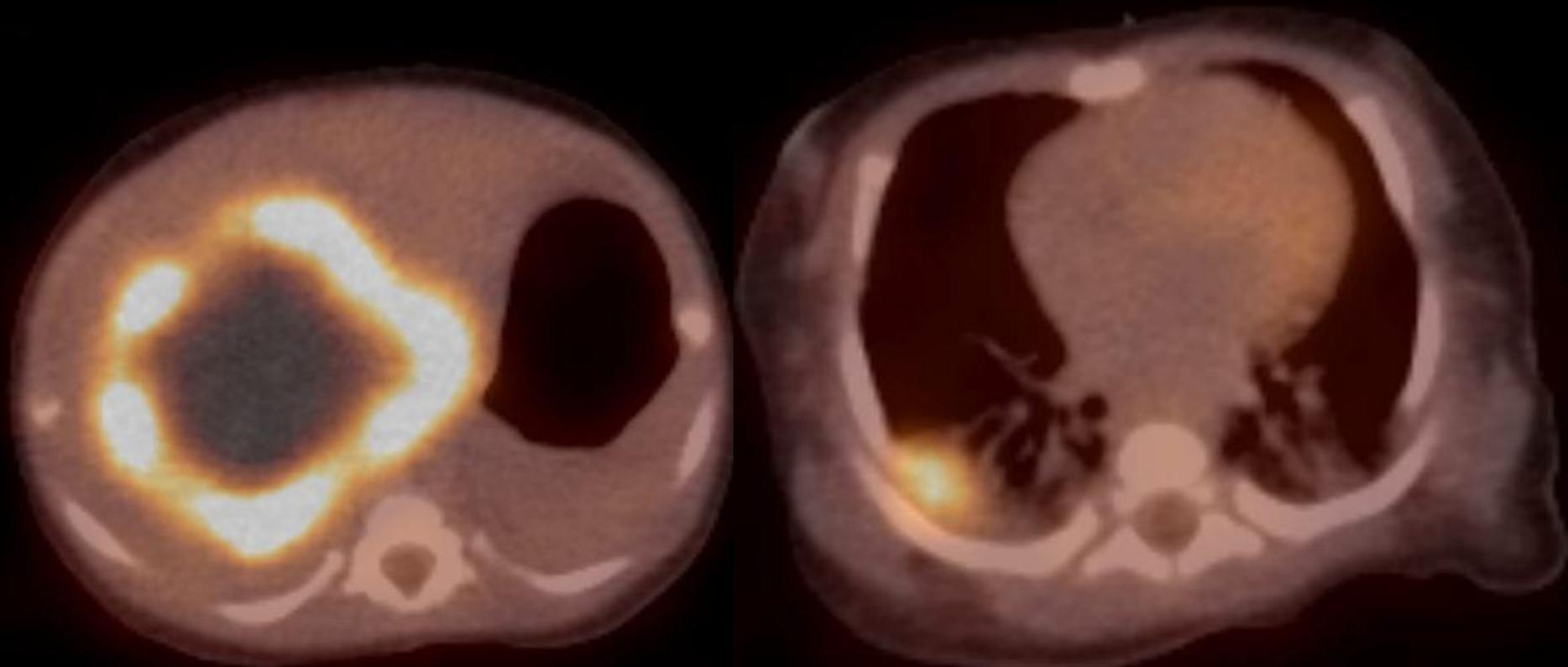
GLABELLA MASS BX 6

Biopsy: Necrosis, with **few malignant cells** seen at periphery of one specimen, insufficient for diagnosis.

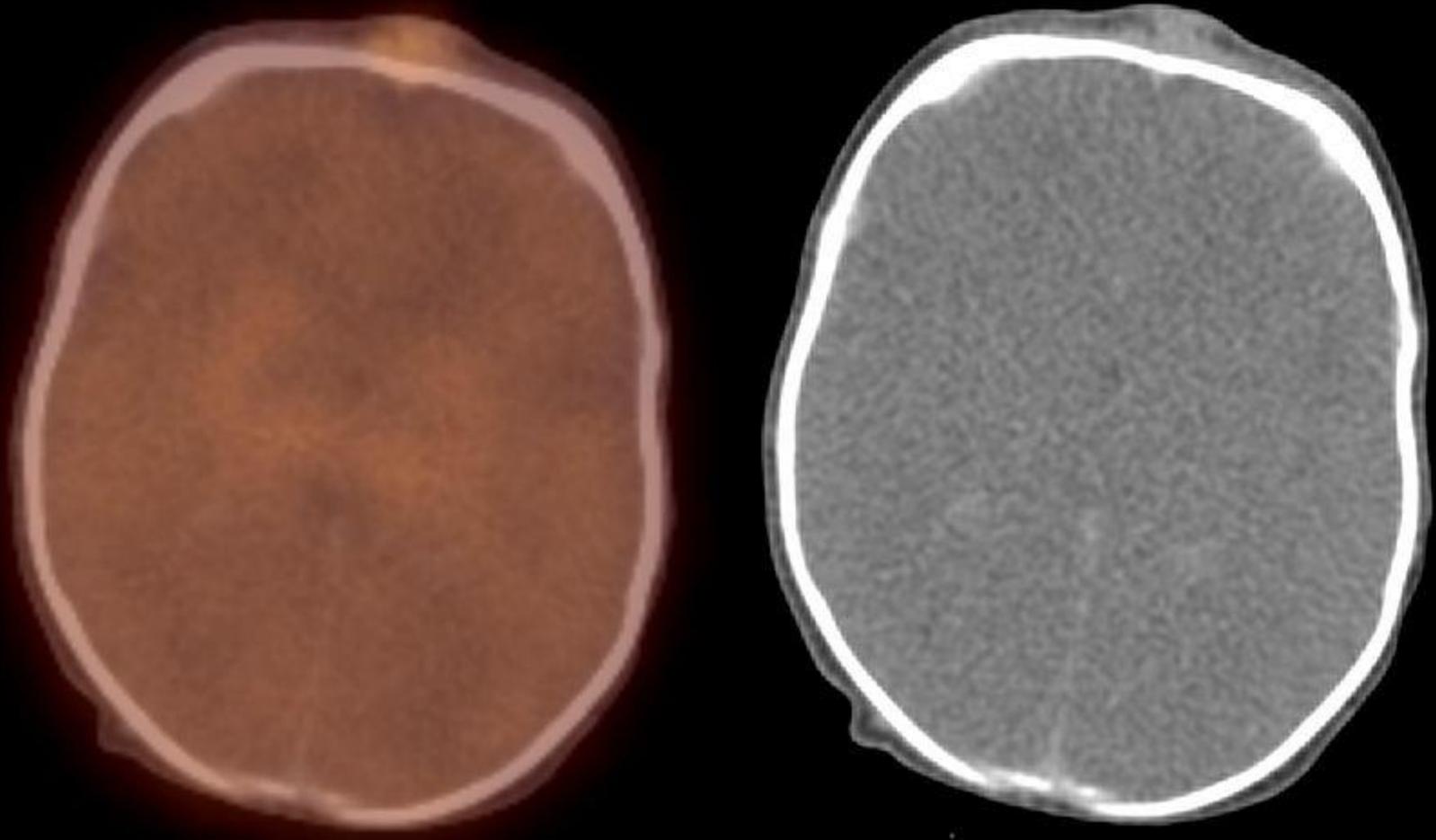


Initial CT Chest/Abd/Pelvis shows peripherally enhancing 6.8 x 6.6 x 5.8 cm liver mass and areas of atelectasis in the lungs with one mass-like area, right lower lobe.





Axial fused PET/CT shows peripherally FDG avid 7 cm liver mass (SUVmax 10.3), small FDG avid lung lesion (SUVmax 4.8).



- Glabellar mass partially resected for tissue diagnosis
- PET/CT shows small amount of residual glabellar mass (SUVmax 3.1)

Diagnosis

- **Choriocarcinoma**
- Liver and lung metastases
- Glabellar cutaneous metastasis
- Gross inspection of placenta was reportedly normal; placenta was not preserved (placental primary is usually small and can go unnoticed on gross inspection)
- Baby found to have elevated beta hCG
- Mother negative for beta hCG

Choriocarcinoma Pathology

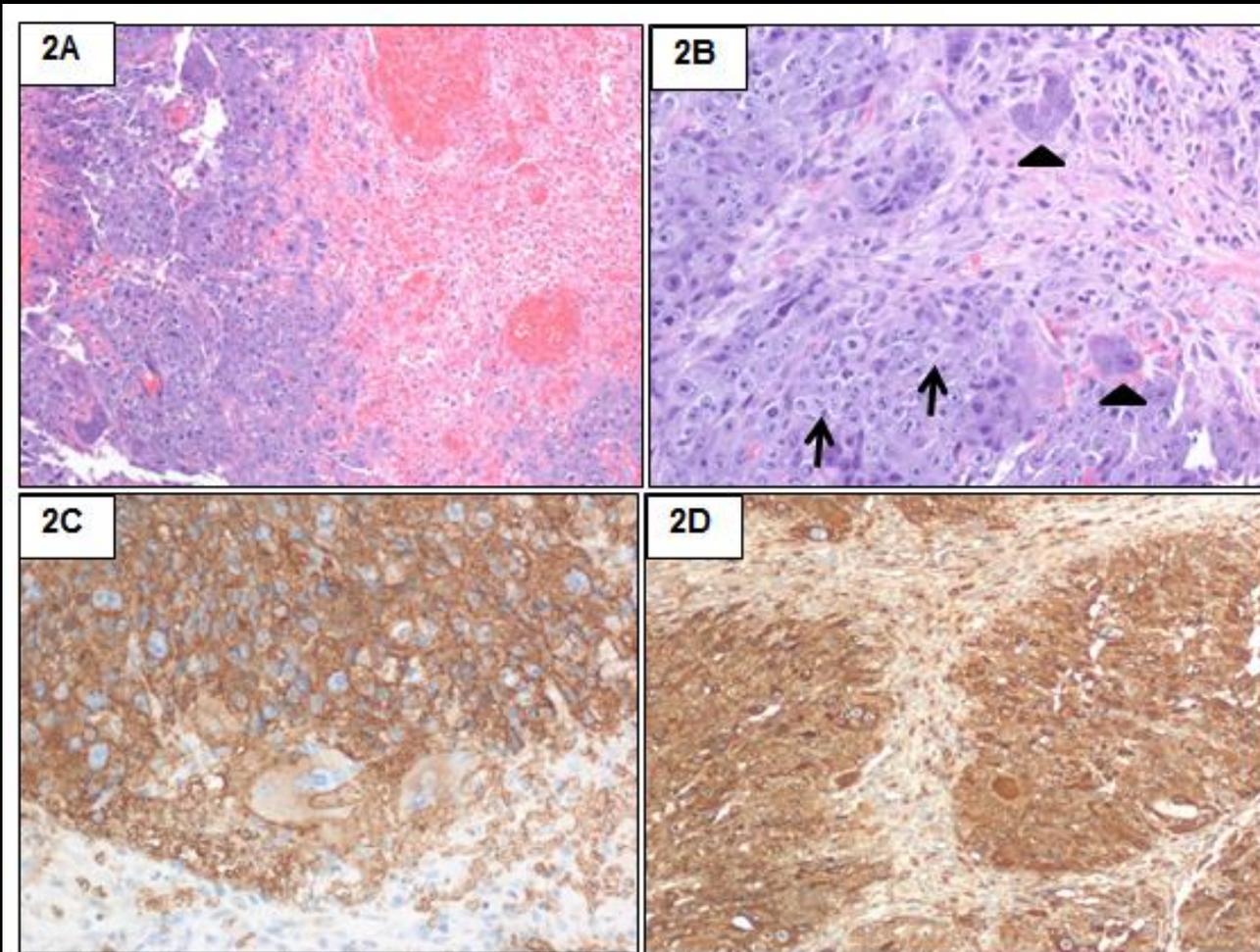


Fig. 2: A – Hematoxylin and eosin (H&E) of the tumor showing a dual population of malignant cells composed of syncytiotrophoblasts and cytotrophoblasts with areas of hemorrhagic necrosis (magnification x 10); B – Higher power view demonstrating bizarre, multinucleated syncytiotrophoblasts (wedges) and cytotrophoblasts with prominent nucleoli and atypical mitotic figures (arrows - x 20); C and D – Neoplastic cells demonstrate marked reactivity with immunohistochemical markers for Glut-1 (C x 20) and β -hCG (D x 10)

Choriocarcinoma

- **Maternal:** Arises from B-hCG secreting placental trophoblastic cells.
 - First solid tumor to be cured with chemotherapy (methotrexate, 1956, Min Chiu Li)
 - Friable, aggressive hemorrhagic metastases to pelvis, distant metastases to lungs, brain, liver.
 - Usually found after molar pregnancy.
 - Occurs in 1 of 50,000 live births.
- **Infantile:** Much less common, thought to be of same origin as maternal, usually concurrent with maternal disease.
 - Metastases usually to the **liver**, although **lung** and **cutaneous** metastases have been described.
 - Highly vascular **rapidly growing** metastases with central necrosis.
 - Treatment: Chemotherapy, delayed surgical resection of mets.
 - Without treatment, **patients usually die within 3 weeks.**

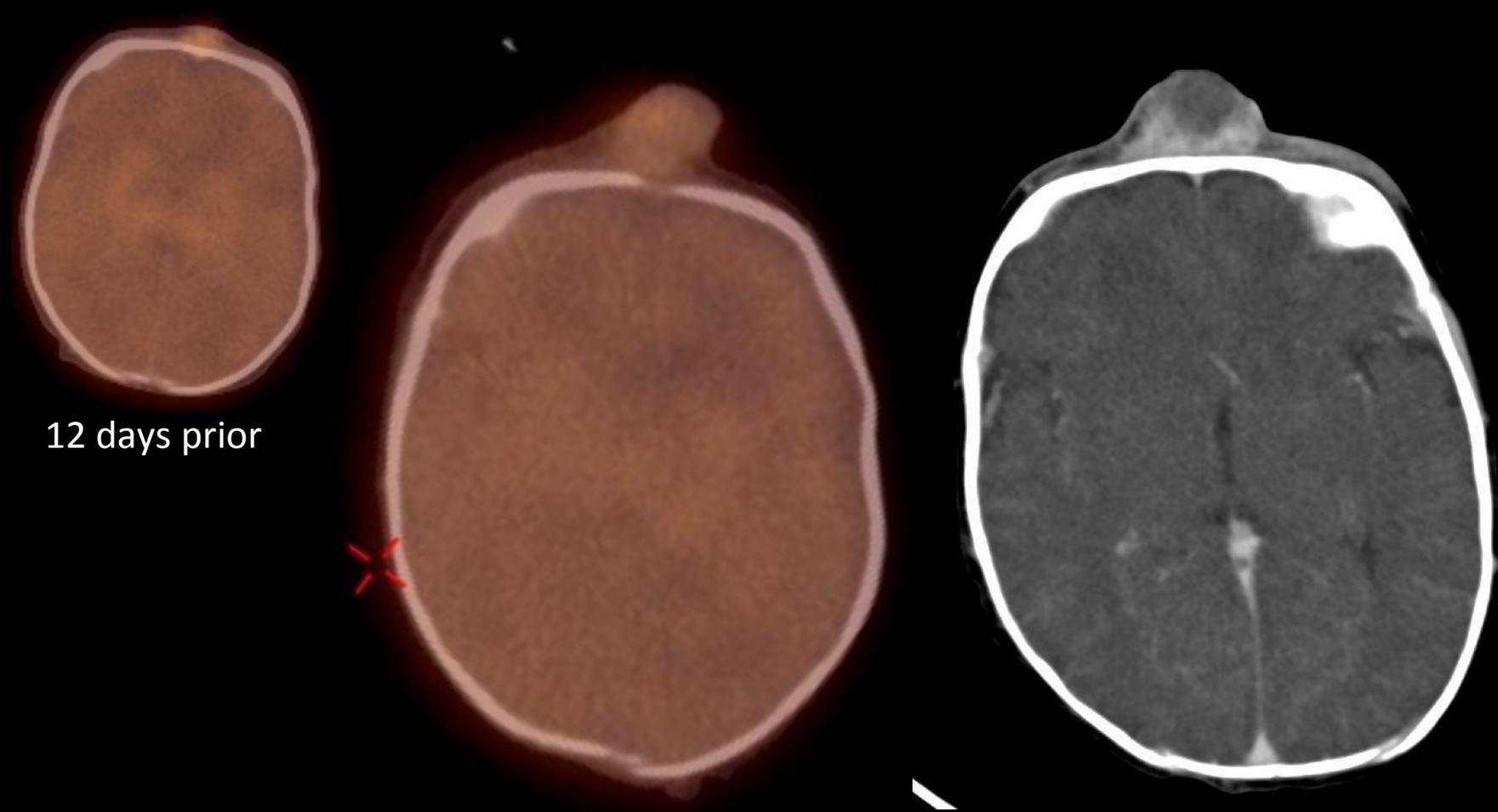


Min Chiu Li

Image courtesy of National Cancer Institute, Wikipedia public domain

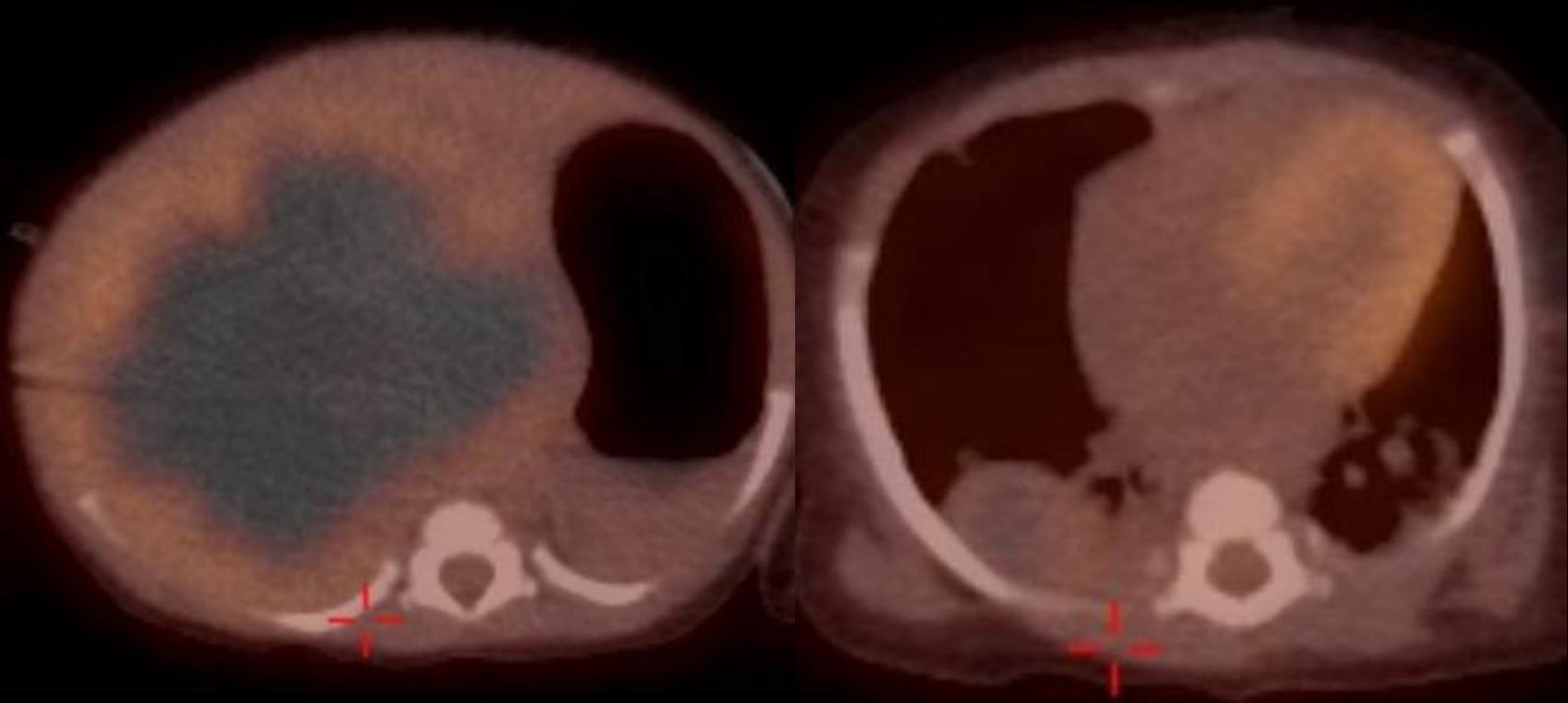
Clinical Course

- Respiratory failure and severe anemia required ICU stay and multiple blood and platelet transfusion
- Rapid improvement shortly after starting chemotherapy



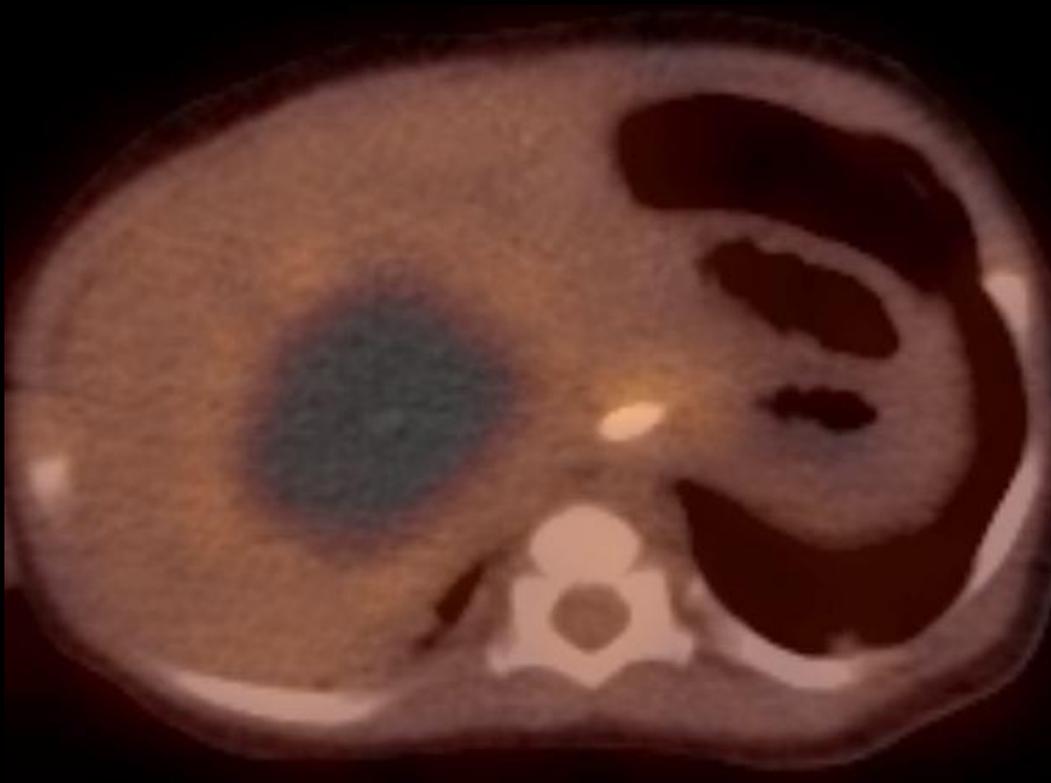
12 days prior

PET **12 days later**: dramatic interval regrowth of glabellar tumor but decreased FDG activity (SUVmax 2.1). Contrast-enhanced CT **14 days later** again demonstrates regrowth and peripheral enhancement of glabellar mass.



PET 12 days later: interval growth but significantly decreased FDG activity of both the liver and lung masses.

3-month follow-up



Images courtesy of Kim Eisenstein, MD, Dermatology, Phoenix Children's

- PET/CT: Liver lesion much smaller, less FDG avid (treatment response).
- Glabellar lesion decreased in size

Treatment

- Choriocarcinoma is treated as a germ cell tumor with resection of all metastases
 - Therapy was initiated per COG protocol AGCT 01P1 (6 cycles cisplatin, etoposide, bleomycin)
 - Right lung lobectomy performed
 - Residual glabellar lesion resected
 - Hepatic lesion cannot be resected fully, therefore patient listed for liver transplant

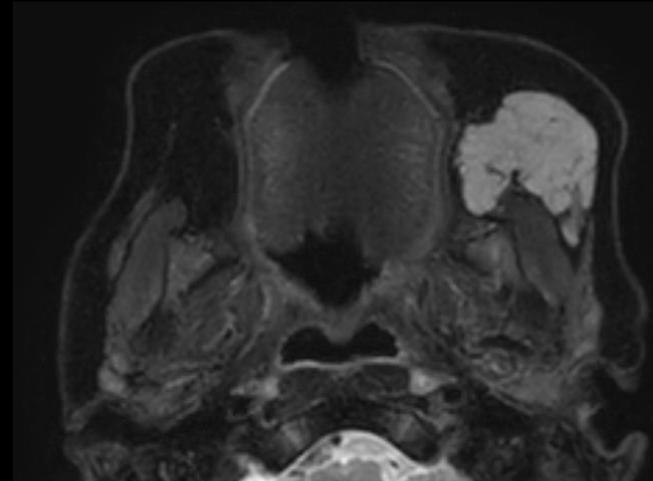
Cutaneous Metastases in Neonates

- Relatively common presentation of malignancy, unlike older children and adults.
- Solitary or multiple varying-size firm, violaceous bluish skin nodules or “blueberry muffin baby”
- From most to least common:
 - Leukemia
 - Langerhans cell histiocytosis
 - Neuroblastoma
 - Rhabdoid tumor
 - Rhabdomyosarcoma
 - Primitive neuroectodermal tumor
 - **Choriocarcinoma**
 - Adrenocortical carcinoma

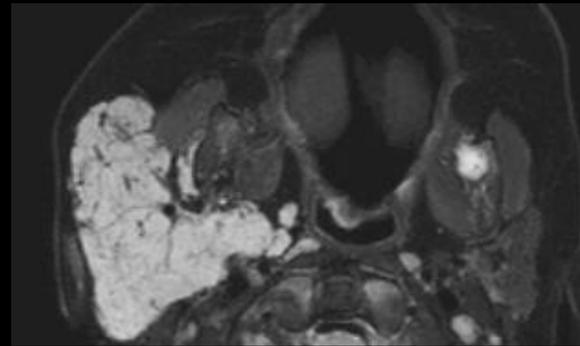


Typical MRI characteristics of untreated infantile hemangioma

- Lobular well-circumscribed solid mass
- Homogeneous T2 hyperintensity is most characteristic feature
- Feeding vessels create flow voids
- Uniform, intense contrast enhancement



3 month old with left cheek infantile hemangioma on axial T2-weighted MRI

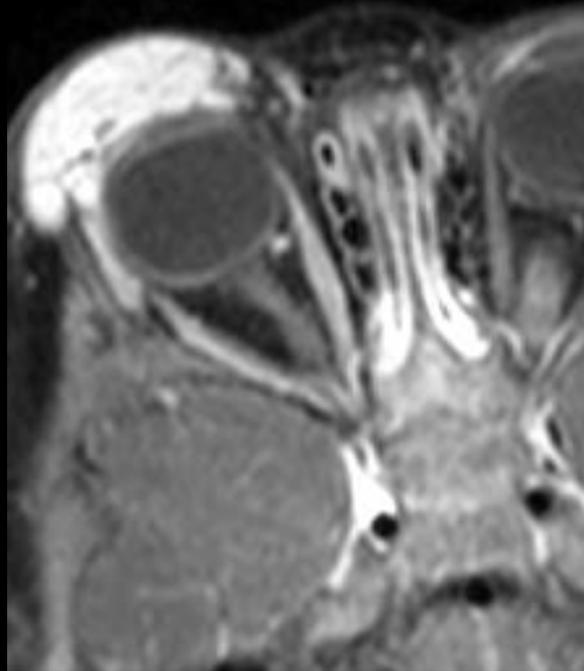


Axial T2: 11 mo old, R parotid hemangioma

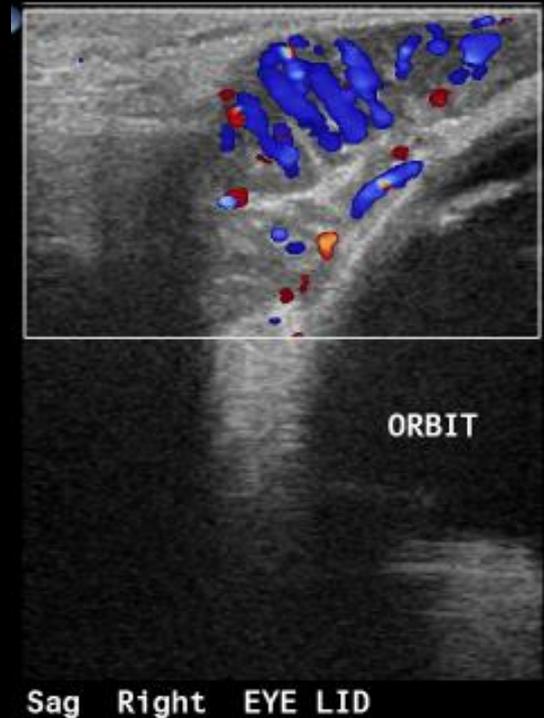
Another infantile hemangioma



Axial T2: 9 mo old with right periorbital hemangioma demonstrating uniform T2 hyperintensity with small flow voids



Axial T1 post-contrast: Intense, uniform contrast enhancement

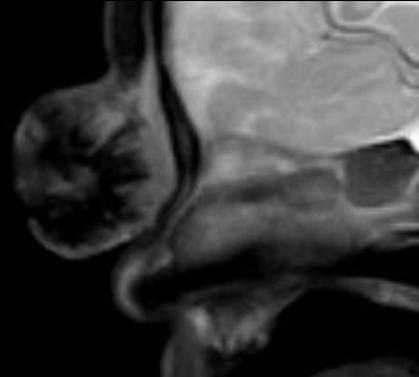


Color Doppler US: Intense arterial and venous color flow throughout the lesion

Typical hemangioma

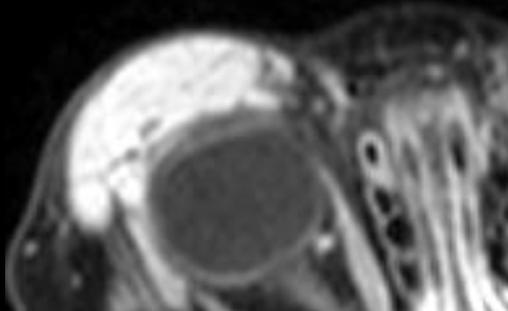
Our case: choriocarcinoma

T2



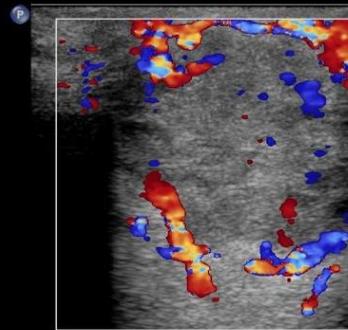
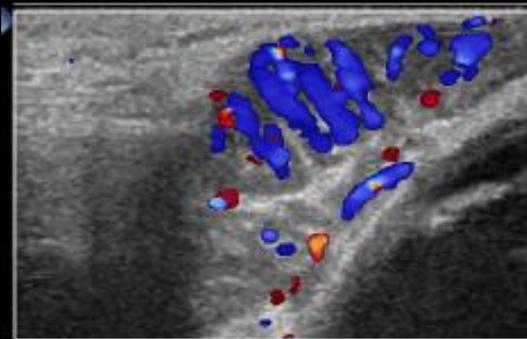
Heterogeneous on T2, no flow voids

T1 post



Peripheral enhancement

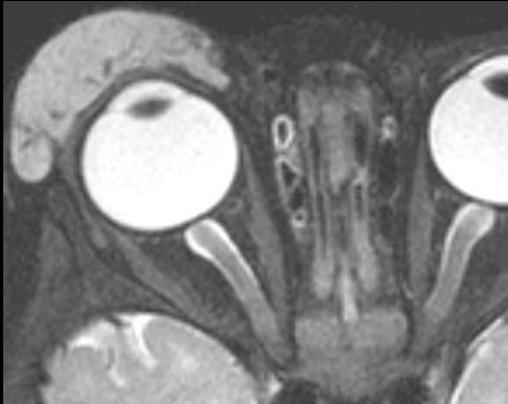
Ultrasound



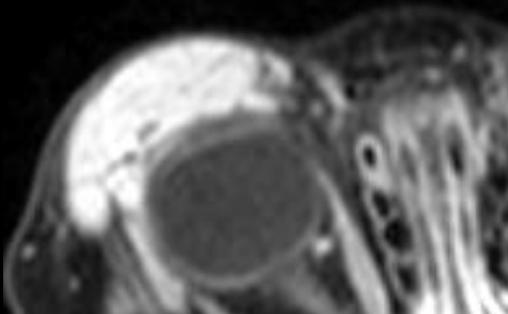
Peripheral hypervascularity, minimal central flow

Typical hemangioma

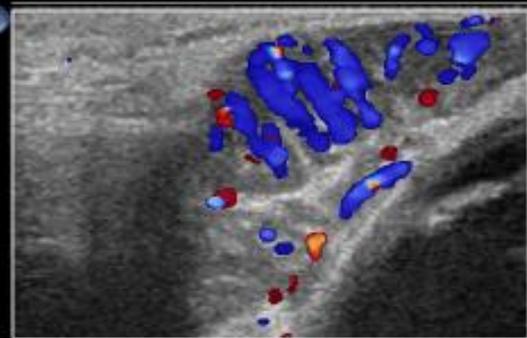
T2



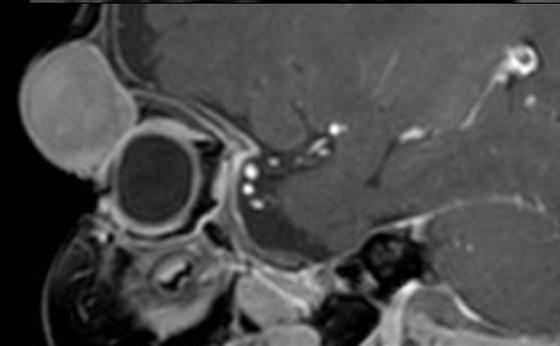
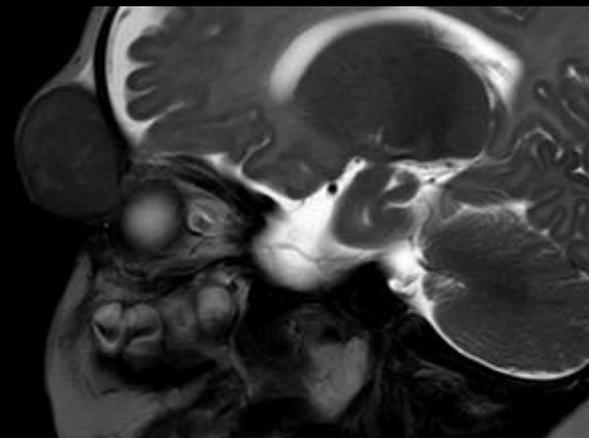
T1 post



Ultrasound



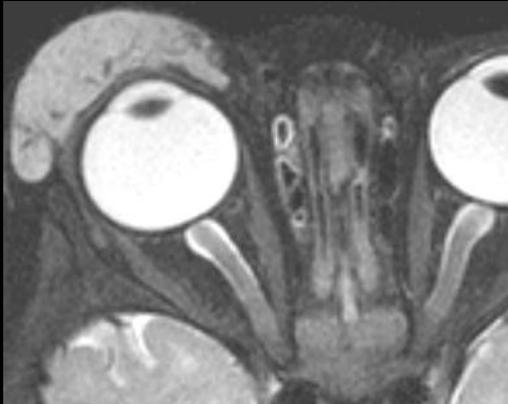
Ewing sarcoma initially diagnosed as infantile hemangioma, 3 mo old



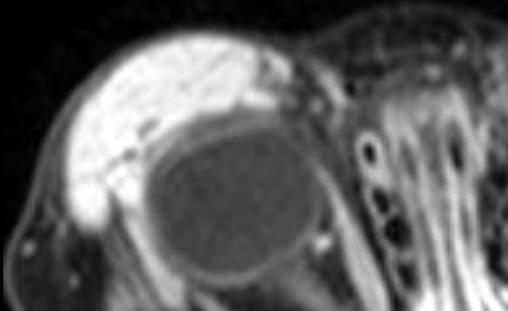
T2 hypointense, less avid contrast enhancement

Typical hemangioma

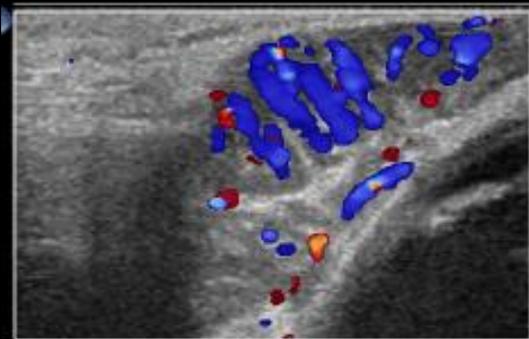
T2



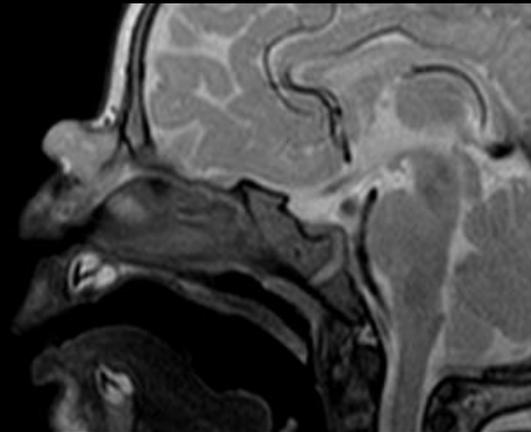
T1 post



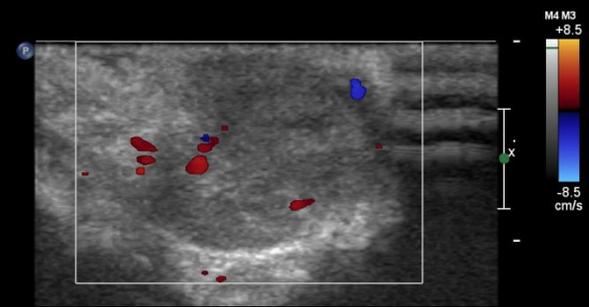
Ultrasound



Nasal glioma also initially diagnosed as infantile hemangioma, 8 mo old



Not as T2 hyperintense, no flow voids, less vascular on ultrasound



Further Discussion on Choriocarcinoma

- **Clinical presentation:** The unusual predilection of infantile malignancies for cutaneous metastases allows their early discovery and creates vivid clinical stigmata not easily forgotten. While relatively common in infancy, such presentations are uncommon in children and adults. The unsightly cutaneous metastasis in our case hastened the diagnosis and life-saving treatment.
- **Known Mimic:** The misdiagnosis of infantile choriocarcinoma for infantile hemangioma has occurred previously, as described in a 2014 case report by Brooks and Nolting also with a solitary cutaneous metastasis on the face.

Further Discussion on Choriocarcinoma

- **Imaging Findings:** Strong, early peripheral enhancement with central necrosis and hemorrhage. These attributes are nonspecific and can mimic an involuting hemangioma or other solid tumors. PET/CT shows marked peripheral FDG avidity; preliminary studies in maternal choriocarcinoma suggest PET/CT is useful in staging and follow-up.
- **Prognosis:** Considered one of the fastest growing tumors, infantile choriocarcinoma classically presents with hepatomegaly, anemia, failure to thrive, and precocious puberty between 0 days and 5 months of life. Chemotherapy and delayed surgical excision of metastases can be curative with long-term survival rates of around 80%.

Conclusion

- Beware the potential of solitary cutaneous metastases to mimic hemangioma both clinically and radiographically
- Typical MRI appearance of infantile hemangioma includes T2 hyperintensity, flow voids, and uniform contrast enhancement
- Atypical features can suggest alternate diagnosis
- Early diagnosis and treatment for infantile choriocarcinoma is critical for survival!
- PET/CT may be useful for staging and follow-up

Sources

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