中文題目:以高免感度肌鈣蛋白(hsTroponin I)即亮點追蹤超音波早期偵測因接受 anthracyclines 治療產生心臟毒性的乳癌病患

英文題目: Action of Sirolimus on a Young Adult with Refractory Vascular Anomalies: A Case Report 作 者:何俊慜<sup>1</sup>, 李欣倫<sup>1</sup>, 林昱廷<sup>2</sup>, 夏和雄<sup>3</sup>,

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## Introduction

Vascular anomalies consist of a heterogenous group of vascular and lymphatic disorders. Most of the vascular anomalies have benign clinical behavior. However, a minority of patients may develop proliferative, destructive, and painful vascular anomalies. Treatment may be tailored by the histopathologic type, location and size of the lesions. Recently, mammalian target of rapamycin (mTOR) inhibitors have been explored as a therapeutic option for complicated vascular anomalies. mTOR activates cell proliferation and angiogenesis via protein synthesis and increased expression of vascular endothelial growth factor (VEGF). This mechanism plays a vital role in the pathogenesis of vascular anomalies. Sirolimus is well-tolerated in pediatric patients with less toxicity. Here, we present a case of refractory cavernous hemangioma in a young female adult treated with Sirolimus for 14 months.

## Case Presentation

This is a 28 year-old female patient with medical history of right hand vascular malformation since birth. She underwent four surgical excisions and twice sclerotherapy for refractory vascular malformation at right thenar eminence since her age of 11. The pathology report of the first surgical specimen was cavernous hemangioma. The latest recurrence was occurred at age 26

Sirolimus was recommended as alternative treatment. She received oral sirolimus at 0.8 mg/m2 per dose twice daily. A month later, she felt her right hand lesions becoming smaller, painless and softer. MRI showed partial response with most of the lesions regressed to smaller size. After 14 months of treatment, the lesions continued to subside

## Discussion

Our patient showed dramatic treatment response of cavernous hemangioma to Sirolimus without experiencing any significant side effect. The effect of Sirolimus is still going on with the lesions continue to shrinkage. The response duration is persisted with this low dose Sirolimus, comparing with surgery and sclerotherapy. The response rate of Sirolimus had been reported around 80-100% with low toxicity in blood function and lipid metabolism. Most of the studies reported its outcome in children. Our study showed same response rate in an adult without obvious toxicity noted to date.

However, we are still concerned the safety of Sirolimus for pregnancy. Adverse events have been observed in animal reproduction studies. Effective contraception should be advised for fertile women

## Conclusion

mTOR inhibitors is a promising medicine in the management of complicated vascular anomalies, namely high response rate and a satisfactory safety profile. Our experience further supports its efficacy on adult.