

Treatment of Aggressive Digital Papillary Adenocarcinoma—Amputation vs. Digit Salvage

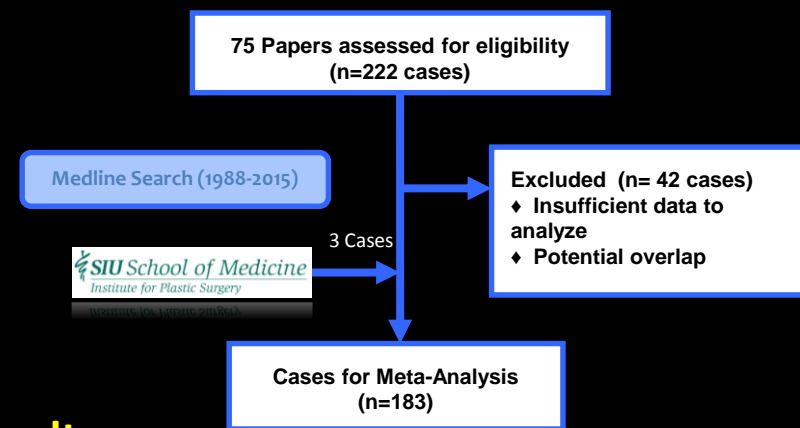
Shaun D. Mendenhall, MD; Ryan W. Schmucker, MD; Jennifer L. Koechle, MPH, CCRP; Steven J. Verhulst, PhD; and Michael W. Neumeister, MD
 Institute for Plastic Surgery, Southern Illinois University School of Medicine, Springfield, IL

Objectives

Aggressive digital papillary adenocarcinoma (ADPA) is a rare tumor of eccrine sweat gland origin with predilection for the hand. Because the rarity of this tumor, published data include only case reports or small case series, making it difficult to define an appropriate treatment algorithm. We performed a meta-analysis of the current world literature in order to compare outcomes between amputation and digit salvage.

Methods

We searched the Medline database to locate all ADPA cases reported in the world literature. Translations were performed as necessary. We included reports of individual variables and excluded reports without case-specific information. The data was compiled and a meta-analysis of treatment methods and outcomes was performed. Descriptive statistics and intergroup comparisons were performed with a p value of <0.05 considered significant.



Results

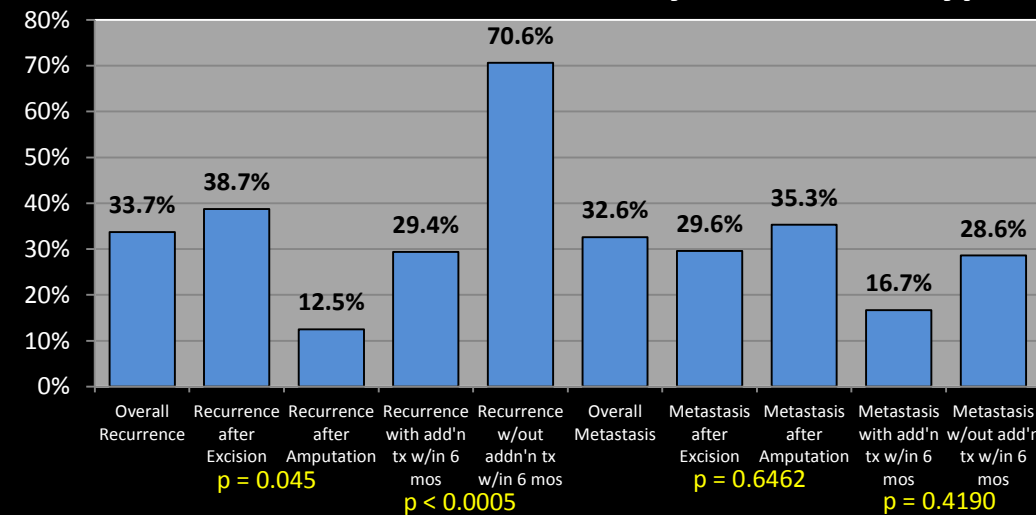
222 cases were mentioned in the literature, 183 were included in the meta-analysis (including 3 of our own patients). Mean patient age was 48 (range 11-85), and 82% were male. Average tumor size was 2 cm and 81% were on the upper extremity. Duration of lesion until diagnosis was 4.8 years, and 4.9% had a history of trauma. Common treatment was excision (88%) vs. amputation (12%). The overall recurrence rate was 33.7%, and overall metastasis rate was 32.6%. Recurrence rate by initial treatment was 38.7% for excision and 12.5% for amputation. Metastasis rate was 29.6% for excision and 35.3% for amputation. Average follow was 44.8 months with 80% of patients were alive and well, 11% living with the disease, 6% dying of the disease, and 3% dying of unrelated causes.



	All	< 25 years old
Age (years)	47.6	17.7
Upper extremity (%)	81.1	27.2
Duration to Dx (years)	4.8	2.1
Tumor Size (cm)	2.0	2.3
Recurrence (%)	33.7	50.0
Metastasis (%)	32.6	42.9
Follow up (years)	3.7	4.8

	Overall	Amputation	Excision
Treatment	99%	11 %	88 %
Recurrence rate	33.7 %	12.5 %	38.7 %
Metastatic rate	32.6%	35.3%	29.6 %

Recurrence and Metastasis by Treatment Type



Discussion

Results of this meta-analysis demonstrate a lower recurrence rate of ADPA when treated with amputation compared to local excision. Treatment modality did not have an impact on metastatic rate. Early definitive treatment (within 6 months) with either complete excision or amputation also conferred a lower recurrence rate than delayed treatment.

The overall metastatic rate of patients in this meta-analysis was 33%, which is higher than previously reported rates of 14% - 26%.^{1,2} Comparing patients with subsequent treatment within 6 months of diagnosis showed a decrease in metastatic rate from 29% to 17%, however this did not reach statistically significant.

This study also demonstrated some unique features of ADPA in younger patients. In 11 cases in patients less than 25 years old, 73% of tumors occurred on the lower extremity, and 55% of tumors appeared on females; both surprising since this tumor has a predilection of the upper extremity of males. ADPA in younger patients was also more aggressive with 50% experiencing recurrence and 43% with metastasis. Amputation may be a better treatment than excision for those who present before age 25.

Conclusions

To our knowledge, the current report is the only meta-analysis of published ADPA cases and the largest series comparing outcomes of amputation vs. digit salvage. Recurrence rate was statistically lower in patients who had amputations (p = 0.045), however, there was no difference in metastatic rates. Due to detrimental impact of amputation on form and function, local excision remains a viable treatment option but patients need to understand the increased risk of recurrence with this treatment. Young patients may benefit more from amputation since at baseline ADPA is more aggressive in this group. Finally, our study also showed that a definitive surgical procedure within 6 months of diagnosis was shown to significantly reduce recurrence. Early diagnosis and complete excision of ADPA will likely lead to improved outcomes that further promote digit salvage.

References

1. Duke W, Sherrod TT, Lupton G. Aggressive digital papillary adenocarcinoma (aggressive digital papillary carcinoma revisited). *Am J Surg Pathol* 2000;24:775-84.
2. Suchak R, Wang WL, Prieto V, et al. Cutaneous Digital Papillary Adenocarcinoma, A Clinicopathologic Study of 31 Cases of a Rare Neoplasm With New Observations. *Am J Surg Pathol* 2012; 36(12): 1883-91.