Case Report

Recurrent Eccrine Poroma with Malignant Transformation and Bony Involvement of the Foot: A Case Report and Review of the Literature

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Objective: *Eccrine poroma is a benign appendage tumor showing either eccrine or apocrine differentiation. However, malignant transformation of this tumor is very rare. The present report demonstrated a case of the eccrine poroma with malignant transformation and its rarity in terms of its high degree of recurrences and its aggressiveness with bony invasion.*
Case Report: *A 67-year old female had a mass on plantar surface of 4th-5th intermetatarsal area of her left foot for 10 years with three recurrent episodes following excisions at local hospital with no pathological report. She was referred to our institute. A recurrent tumor was removed again with pathological findings as eccrine poroma with incomplete excision; however, the patient had a failure to follow-up. Four years later, she was back in our center again with 5th episode of recurrent tumor which was marginally excised with pathological findings revealing a malignant transformation of eccrine poroma with close resected margins. Unfortunately, the patient failed to follow-up again. Eventually, four years later, she returned to our institute with 6th recurrent episode of tumor. Incisional biopsy was performed with pathological findings as appendage tumor with eccrine differentiation. Wide resection including removal of 5th metatarsal head-and-neck was performed due to aggressive behaviors of this tumor; particularly; history of malignant findings and multiple recurrences, significantly bony invasion. Final pathological findings were malignant eccrine poroma with close resected margin. There were no metastases in evidence from all investigations. Local irradiation with the aim of eradication of microscopically residual tumor was initiated.*
Conclusion: *Malignant transformation is a rare occurrence of eccrine poroma. It should be initially included in differential diagnoses, especially in a patient with long standing foot lesion or history of recurrent masses. Wide excision is recommended as basis treatment, especially in a patient with high likelihood of recurrences or positive malignant transformation.*

Keywords: *Malignant eccrine poroma, Porocarcinoma, Eccrine poroma, Recurrence, Malignant transformation, Foot*

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Eccrine poroma is an adnexal neoplasm with terminal ductal differentiation that shows either eccrine or apocrine differentiation. It usually present as solitary tumor on the sole or the sides of foot. However, it can be seen in any cutaneous location. Most poroma arise in middle age with no sex predilection. However, the malignant transformation of this tumor is very rare[1,2]. The tumor develops gradually and can be present for a number of years without obvious transformation. The recurrence rate is about 20%[3]. The metastases of these eccrine tumors become visible first in regional lymph nodes while the lungs are the most common organ affected by hematogenous metastases. Unsuccessfully identifying its clinical and morphologic features may lead to overlooking this diagnosis resulting in inadequate treatment[4]. The present report is to demonstrate a case of an eccrine poroma with its rare malignant transformation in terms of its high degree of recurrence and its aggressiveness with bony invasion.

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Also included are the clinical and histopathological findings, the treatment course and the results. A concise review of the literature, including the histopathological characteristics and management, is also presented. The permission of the patient was obtained for this report.

Case Report

A 67-year old female had a mass on the plantar surface of the 4th-5th intermetatarsal area of her left foot for 10 years that attained a size of 40 x 40 x 20 mm. While it had been developing for the last year, it was removed 3 times due to recurrent events, at a local hospital. There was no pathology report from that hospital. The tumor recurred 3 months after its last excision. The patient was then referred to our institute. A recurrent tumor was removed again; the pathology report was an eccrine poroma with incomplete excision. The patient failed to follow-up as recommended. Four years later, she was back in our center again with the 5th episode of this recurrent tumor. At this time, a rubbery mass was located at the previous surgical area with a size of 38 x 27 x 8 mm. There was no evidence of bony involvement from the radiographic findings. A recurrent tumor was marginally excised and the pathology findings revealed a malignant transformation of an eccrine poroma with close resected margins (1 mm) and no angiolymphatic or perineural invasion. Unfortunately, a patient failed to follow-up again because she could walk without pain after the operation. Once again, four years later, she returned to our foot clinic with the 6th recurrent episode of this tumor attaining a size of 35 x 25 x 12 mm. The tumor had the presentation of a firm consistency with mild mobility (Fig. 1). No regional lymph node enlargement was found and other findings were within normal limits on physical examination. The complete investigation consisted of the plain radiographs, magnetic resonance imaging (MRI), bone scintigraphy of whole body, and chest computed tomography (CT). The plain radiographs revealed a widening of the distal portion of the 4th-5th intermetatarsal area, significantly bony erosion and osteopenic changes of the 5th metatarsal's head and neck (Fig. 2). The MRI delineated a lobulated, ill-defined mass at the plantar surface between the interrosseous space of the 4th and 5th metatarsal bones. This is demonstrated by isosignal intensity on T1W with involvement of the 5th metatarsal head (Fig. 3A), the heterogeneous hyper signal intensity with central isosignal intensity portion as shown on T2W image (Fig. 3B). After contrast administration, this mass shows heterogeneous moderate degree enhancement (Fig. 3C, 3D). The bone scintigraphy of

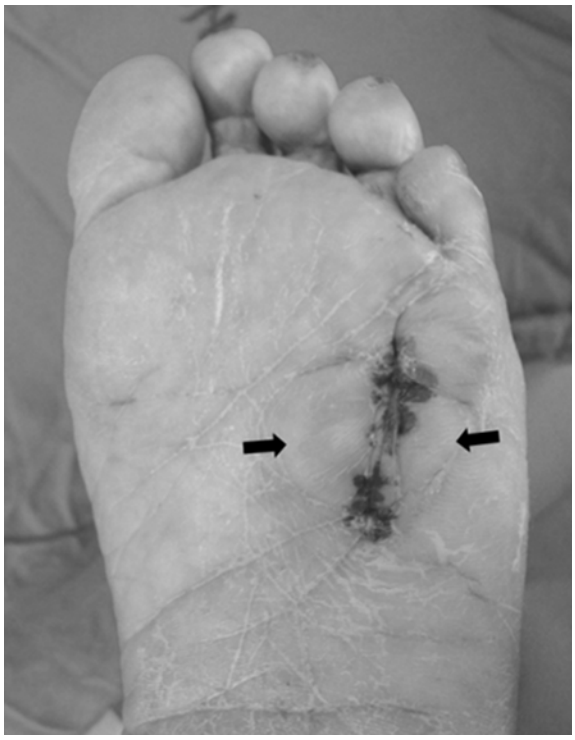

Fig. 1 The gross appearances of the lesion at the plantar surface of the 4th-5th intermetatarsal area are shown (black arrow) with a planned longitudinal incision as marked with a surgical pen marking

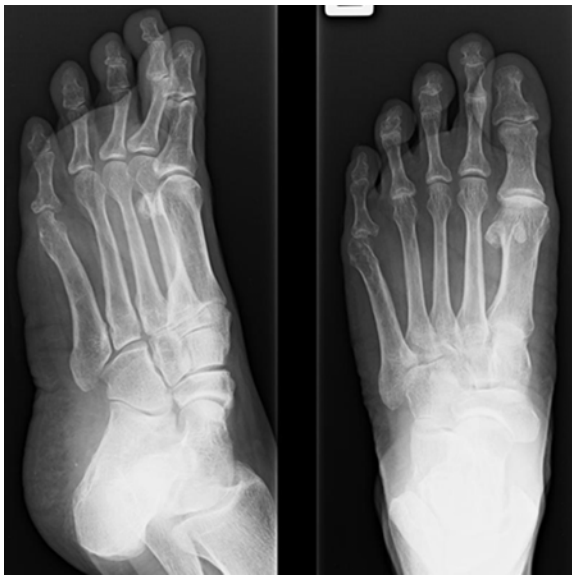

Fig. 2 The anteroposterior and oblique radiographs show the widening of distal part of the 4th-5th intermetatarsal area, significantly bony erosion and osteopenic changes of the 5th metatarsal's head and neck

the whole body showed an increased uptake at the 4th-5th intermetatarsal area. The results of the chest computed tomography (CT) was negative. After all of the investigations were completed, an incisional biopsy was performed. The pathological findings revealed an appendage tumor with eccrine differentiation. Histologic evidence of invasion could not be evaluated due to limited tissue material. Although the pathological findings did not show the actual characteristics of a malignant eccrine poroma, there were various clues to the aggressive behaviors of this tumor as evidenced with a history of malignancy and multiple recurrences, significant bony invasion and an unusual epithelial proliferation found at this time. After the discussion between the physicians, the patient and her relatives, an aggressive resection of the tumor was decided upon as the definitive treatment of this recurrent episode. The wide resection performed included the removal of the 5th metatarsal's head and neck (Fig. 4,5). There were no signs of involvement of the proximal phalange of 5th toe from the gross examination in the operative field. The final pathological findings showed a malignant eccrine poroma with close resected margins (1 mm) (Fig. 6-9).

After the consensus of a foot surgeon (CA), a pathologist (JK) and a radiologist (WA), including the counseling of the patient and her relatives, the authors decided to treat the patient with local irradiation. The goal was for the eradication of the microscopically residual tumor at the previously tumorous area following the complete healing of the surgical wound at three weeks postoperatively. So far as at least 2 months after the completed treatments, there were no signs of recurrence and wound complications. She could walk properly with no pain using a modified shoe.

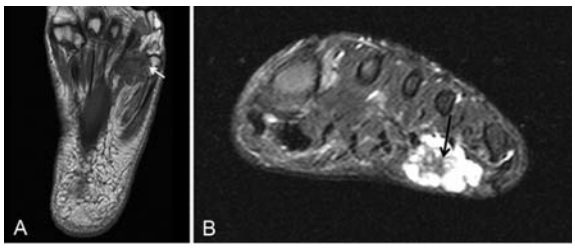

Fig. 3 A) T1W image shows a lobulated, ill-defined mass at the plantar surface between the interrosseous space of the 4th and 5th metatarsal bones which demonstrates isosignal intensity with involvement of the 5th metatarsal head (white arrow) B) T2W image shows heterogeneous hypersignal intensity with central isosignal intensity (black arrow)

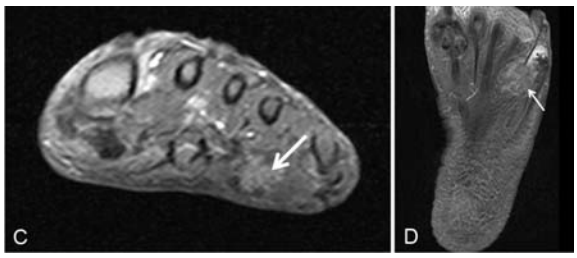

Fig. 3 (C-D) After contrast administration, this mass shows heterogeneous, moderate degree enhancement (white arrow)

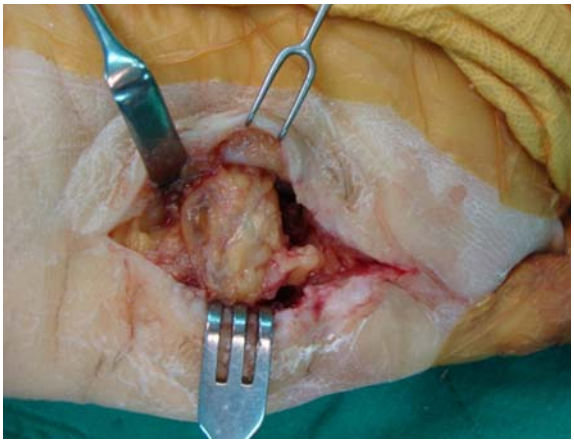

Fig. 4 The intraoperative findings show the gross appearance of the tumor

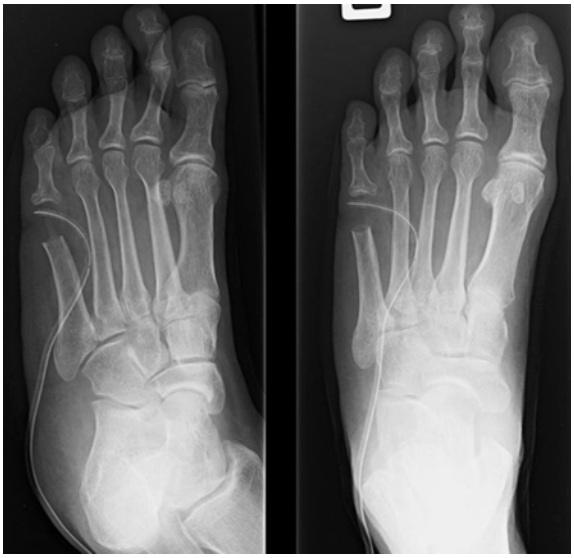

Fig. 5 The postoperative radiographs after the wide excision and removal of 5th metatarsal's head and neck with a postoperative drainage tube

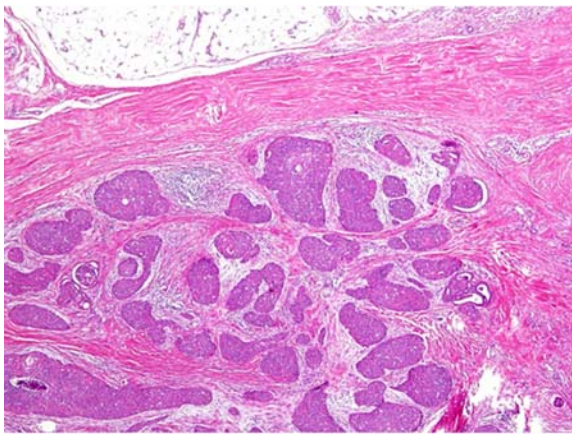

Fig. 6 The tumor is asymmetric and composed of lobules of polygonal tumor cells, infiltrating the subcutaneous tissue

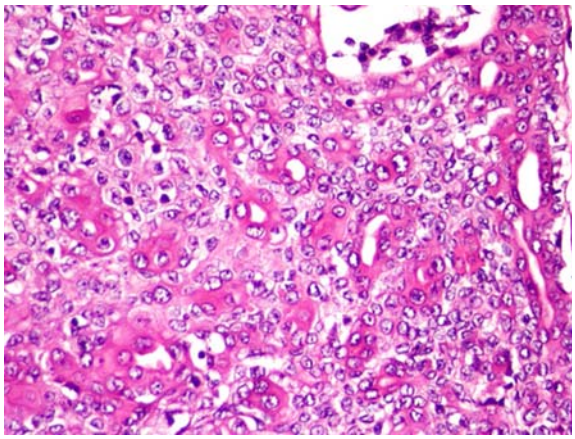

Fig. 8 The tumor reveals ductal differentiation characterized by ducts lined by cuticular eosinophilic material

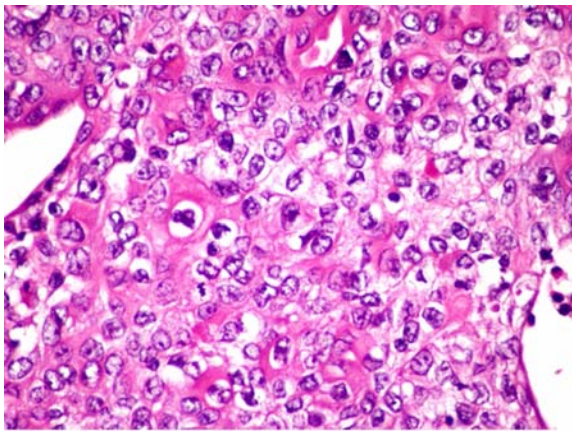

Fig. 7 The malignant cells have large, hyperchromatic, irregularly shaped nuclei and clear cytoplasm (rich in glycogen). Occasional mitoses are seen

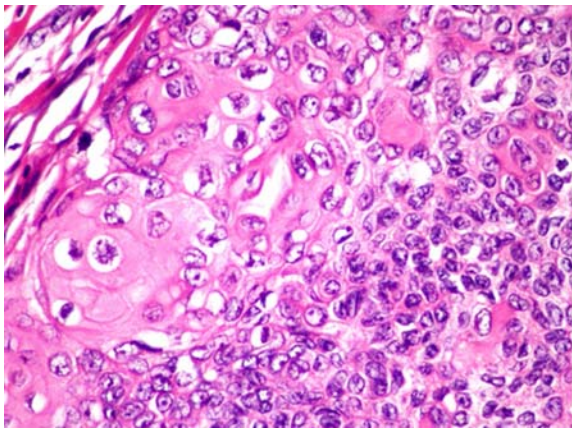

Fig. 9 The tumor shows scattered foci of squamous differentiation characterized by keratinized cytoplasm and intercellular bridges

Discussion

Eccrine poroma is a benign sweat gland tumor which derives from the intraepidermal segment of the eccrine glands. Pinkus et al proposed the first report in 1956[5]. A malignant variant was reported in 1963[6]. This tumor represents 0.1% of all primary skin tumors[4]. Eccrine poroma frequently arises in middle-aged or elderly people. There is no gender predilection[4]. Eccrine poroma regularly occurs as a single, gradual growing, soft to firm, non-painful, skin surface color to erythematous or brown plagues or nodules[7]. It might present with a sessile or broadly pedunculated pedestal[7]. Most lesions are well confined, normally enclosed by a shallow edge. Some lesion may be fairly circumscribed. The size arrays from a few millimeters to less than 2 cm in width[4]. However, it may be larger in cases of a presentation with a long duration. Extremities were the most common sites of involvement, especially in the hands, fingers, soles, or the sides of the feet[4,7]. Most tumors commonly exist as asymptomatic and have been indolent for a long period before the patient seeks a physician for treatment. The patients may present with bleeding or ulceration at this tumor after minimal trauma[4]. It is sometimes diagnosed incorrectly at the initial presentation, and the tumor is simply mistaken as a pyogenic granuloma, an amelanotic melanoma, a basal cell carcinoma, a squamous cell carcinoma, a basalioma, a leiomyosarcoma or a seborrheic keratosis[4]. The sporadic subtype presenting as a pigmented

eccrine poroma that contains melanocytes may mimic a melanoma[8,9]. The definitive diagnosis is essentially retrieved with histological examination. Immunohistochemical investigations may be critical in differentiating this tumor from other mimicking tumors.

Even though eccrine poroma is commonly benign in nature, the recurrence of this tumor and its variants were usually reported after incomplete excision[4]. For the malignant entity, it normally arises from its origination or de novo, but some develop from a long-standing eccrine poroma[9]. The clues for the suspicion of the malignant form are that they are large in size, fast growing, currently increasing in size, present with bleeding, pain or irritation, etc[10]. However, malignant eccrine poroma encompass less than 0.01% of all dermatological biopsy specimens[11]. Moreover, the eccrine poroma and its variants with malignant transformation are incredibly rare[1,2]. This malignant tumor may present with an aggressive course, with wide spread metastases and death[4,12]. There are 20-30% of cases with a delayed presentation and delayed diagnosis including cutaneous metastasis or regional lymph node involvement at the initial diagnosis[12-14]. The worst prognosis was reported with the relationship of regional nodal involvement as a 65% mortality[12]. Solid organ metastases are observed in 10% of cases, lymph node metastases in 20% of cases, and local recurrence in 20% of cases[10,15].

The difficulty in the detection of the malignant variants or the transformation of eccrine poroma may lead to late diagnosis and a poorer prognosis if there is regional lymph node involvement or other metastases. Therefore, the early recognition and appropriate treatment of an eccrine poroma, at the initial presentation to the physician, are essential[4]. The complete resection of the eccrine poroma with histological inspection is necessary to verify the diagnosis and to rule out the existence of dysplastic or malignant variants[4]. Eccrine poroma can recur after inadequate resection. For the treatment of the malignant eccrine poroma or eccrine porocarcinoma, a wide excision is the procedure of choice[13,16]. The total resection of this tumor might be curative and helps to prevent further recurrence[16]. However, the patients with dysplastic changes or malignant forms require regular follow-up to assure early identification of recurrence and metastasis.

In the present report, the patient presented with a long history of the plantar lesion with multiple episodes of a recurrent mass. There were many issues that might explain and be involved in the phenomenon of this case. First of all, the first 3 surgical excisions were performed by a physician in a local hospital with no identification by pathological report. It was possible that some physicians may not consider the differential diagnosis of an eccrine poroma or other appendage tumors at this area as mentioned in the discussion above. One should pay attention to the importance of the histological investigation of tumors in this area, especially, in a recurrent lesion. The present study proposes an incisional biopsy at the initial presentation if there are clues or a history of a malignant nature or possible malignant transformation. The definitive treatment is decided upon after identification by the pathological findings and other completed definitive investigations. Second, the pathological examination of this case showed benign findings with incomplete removal and the malignant transformation with close resected margins at the first and second excisions in our center, respectively. However, the patient's failure to follow-up both times after the excision significantly affected the outcome. This point emphasizes that a physician should advise their patients to appreciate the importance of regular follow-up. This is crucial for a patient with a high risk of recurrence or malignant transformation as presented in this case. Third, the present study reports that the definitive treatment with a wide resection and the 5th metatarsal head and neck removal, as in this case. This treatment is the treatment of choice for patients with tumors exhibiting the aggressive behavior as evidenced with multiple recurrences with significant bony involvement. At this time, secondary to the pathological report that revealed close resected margins (1 mm), the authors decided to treat the patient with adjuvant radiotherapy with the aim of the elimination of the microscopically residual tumor. This rational treatment was in accord to the benefit of patients with local irradiation for the goal of assuring the patient disease-free[17,18].

Conclusion

The present report proposes a case of the eccrine poroma with malignant transformation and its scarcity in terms of its high degree of recurrence and its aggressiveness with significant bony invasion. This tumor should be initially included in the differential diagnoses, especially in a patient with a long duration of a foot lesion or a history of a recurrent mass. After definitive diagnosis from the incisional biopsy with pathological examination and complete investigation, a wide excision is recommended as the mainstay treatment, particularly in cases with a high probability

of recurrence or evidence of malignant transformation.

Potential conflicts of interest

None.

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รายงานเนื้องอกเอ็คครายน์โฟโรมาที่มีการเป็นซ้ำและเปลี่ยนสภาพเป็นเนื้องอกมะเร็งของบริเวณเท้า

ชญานิน อ่างทอง, จุฑาทิพย์ คินทรักษ์, ศุภกิจ คณิตเนตร, วิรณา อ่างทอง

วัตถุประสงค์*: เอ็คครายน์โฟโรมาเป็นเนื้องอกชนิดธรรมดา (ไม่ใช่มะเร็ง) ที่มีการพัฒนาจากส่วนของอะโพรครายน์ อย่างไรก็ตาม การเปลี่ยนสภาพเป็นเนื้องอกมะเร็งนั้นพบได้น้อยมาก รายงานฉบับนี้ได้บอกถึงผู้ป่วยที่มีเอ็คครายน์โฟโรมาที่เริ่มต้นแบบเนื้องอกชนิดธรรมดา และมีการเปลี่ยนสภาพเป็นเนื้องอกมะเร็งในเวลาต่อมาโดยเสนอแง่มุมต่างๆ ของเนื้องอกชนิดนี้ที่พบได้ยากได้แก่ การเป็นซ้ำหลายครั้ง และความรุนแรงในการทำลายกระดูกข้างเคียง*

รายงานผู้ป่วย*: ผู้ป่วยหญิงอายุ 67 ปีมีก้อนที่ฝ่าเท้าระหว่างกระดูกเมตาทาร์ซอลที่ 4-5 มาเป็นเวลา 10 ปี โดยเป็นซ้ำมาแล้ว 3 ครั้ง หลังจากรับการผ่าตัดที่โรงพยาบาลชุมชนโดยไม่มีการรายงานผลทางพยาธิวิทยาหลังผ่าตัด ผู้ป่วยได้รับการส่งมาที่สถาบันนี้เนื่องจากการเป็นซ้ำครั้งที่ 4 และได้รับการผ่าตัดอีกครั้งผลทางพยาธิวิทยาแสดงถึงเนื้องอกเอ็คครายน์โฟโรมาแบบชนิดธรรมดาโดยเลาะออกไม่สมบูรณ์ อย่างไรก็ตามผู้ป่วยได้ขาดการติดต่อไปและกลับมาอีกครั้งช่วง 4 ปีต่อมาด้วยการเป็นซ้ำครั้งที่ 5 โดยได้รับการตัดออกอีกครั้ง ผลทางพยาธิวิทยาแสดงถึงเอ็คครายน์โฟโรมาที่มีการเปลี่ยนสภาพเป็นเนื้องอกมะเร็งแล้ว โดยขอบเขตการตัดออกใกล้เนื้อเยื่อปกติค่อนข้างมาก ผู้ป่วยได้ขาดการติดต่อไปอีกครั้ง สี่ปีต่อมาผู้ป่วยกลับมาอีกครั้งด้วยการเป็นซ้ำครั้งที่ 6 ในครั้งนี้เนื่องจากลักษณะการคุกคามรุนแรงของเนื้องอกนี้ได้แก่ ประวัติเดิมที่มีการเปลี่ยนสภาพเป็นเนื้องอกมะเร็งและการเป็นซ้ำหลายครั้ง, การลุกลามต่อกระดูกเมตาทาร์ซอลข้างเคียง ผู้ป่วยจึงได้รับการผ่าตัดเนื้องอกออกแบบกว้าง ร่วมกับการตัดกระดูกเมตาทาร์ซอลที่ 5 ส่วนปลายซึ่งได้รับการลุกลามจากเนื้องอกดังกล่าวออกไปด้วย ผลทางพยาธิวิทยาครั้งสุดท้ายเป็นเอ็คครายน์โฟโรมาที่เป็นชนิดมะเร็งโดยขอบเขตการตัดออกใกล้เนื้อเยื่อปกติค่อนข้างมาก ผู้ป่วยได้รับการฉายรังสีเฉพาะที่ต่อไปเพื่อกำจัดเนื้องอกที่ขนาดเล็กมากแบบตาเปล่าไม่สามารถมองเห็นได้ ซึ่งอาจหลงเหลืออยู่ โดยได้ทำการตรวจค้นเพิ่มเติมร่วมด้วย และไม่พบว่ามีการกระจายของมะเร็งไปที่ส่วนอื่นของร่างกาย*

สรุป*: การเปลี่ยนสภาพเป็นเนื้องอกมะเร็งของเอ็คครายน์โฟโรมาแบบชนิดธรรมดาพบได้น้อยมาก ภาวะนี้ควรได้รับการตระหนักถึงสำหรับการวินิจฉัยแยกโรคต่างๆ ในผู้ป่วยที่มาด้วยการมีก้อนที่ฝ่าเท้าเป็นเวลานาน หรือก้อนเกิดขึ้นซ้ำหลายครั้ง การผ่าตัดเนื้องอกออกแบบกว้างเป็นการผ่าตัดที่เป็นหลักในการรักษาเนื้องอกลักษณะนี้ โดยเฉพาะอย่างยิ่งในผู้ป่วยที่มีความเป็นไปได้สูงที่จะมีการเป็นซ้ำหรือการเปลี่ยนสภาพเป็นเนื้องอกมะเร็ง*