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下顎骨骨性肉瘤之病例報告

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摘 要

頭頸部的惡性腫瘤中，肉瘤(Sarcoma)所佔的比例僅在百分之壹左右；而頭頸部肉瘤中，骨性肉瘤(Osteogenic sarcoma)僅爲少數。本篇報告壹例下顎骨骨性肉瘤病患；病患爲 25 歲女性，求診時主訴爲下顎骨有如石頭般之硬性腫塊，腫塊部位包含整個下頷部及其兩側。病患曾壹年半前求診其他醫院並接受三次切除手術；診斷報告分別爲下顎骨纖維性發育不良病，及下顎骨骨性肉瘤。經臨床理學檢查及影像學檢查後，診斷爲下顎骨聯合部及兩側骨體之骨性肉瘤。病患接受兩側肩胛舌骨肌上淋巴結分離術(Supraomohyoid neck lymph node dissection)、塊狀下顎骨切除術、口內腫瘤廣泛切除術、及小腿腓骨骨頭皮膚游離皮瓣重建手術。術後恢復良好；病患於術後兩個月再接受化學治療。因下顎骨骨性肉瘤爲罕見之病例，故本篇特提出此病患之病症、臨床理學檢查、影像學檢查、治療方式、及追蹤情之情況以提供參考。

關鍵語：下顎骨，骨性肉瘤。

引 言

骨性肉瘤是由一群可以製造類骨質(Osteoid)及不成熟骨頭之間質細胞(Mesenchymal cell)所組成之惡性腫瘤。骨性肉瘤可分為低度中央骨性肉瘤(Low grade central osteogenic sarcoma)、骨膜外骨性肉瘤(Parosteoal osteogenic sarcoma)、骨膜骨性肉瘤(Periosteal osteogenic sarcoma)、及高度骨性肉瘤(High grade osteogenic sarcoma)[1-3]。骨性肉瘤為肉瘤之一種，而肉瘤為較少見之惡性腫瘤，在頭頸部的惡性腫瘤中，肉瘤所佔的比例依各文獻紀錄僅在百分之壹左右；而頭頸部肉瘤中，骨性肉瘤僅為少數[4-7]；本篇報告即報告壹例下顎骨高度骨性肉瘤之病患。

骨性肉瘤為僅次於多發性骨髓瘤(Multiple Myeloma)之骨惡性腫瘤，且約佔全身惡性腫瘤中 0.2%左右，男女發生比例約為 59%比 41%，好發於 10-19 歲[8-16]。而發生於下顎骨之骨性肉瘤為極少數[17,18]，故本文提出此下顎骨高度骨性肉瘤病例之病患病史，理學檢查，放射線檢查，手術方法，病理報告，及其術後追蹤之記錄，再配合文獻報告，以供參考。

病 例

患者求診時為二十五歲女性，於民國 91 年 10 月 3 日來本院口腔顎面外科初診，求診時主訴為整個下顎骨如石頭般硬度、有一大約 10 cm ×6 cm×6 cm 之腫塊，並感覺此腫塊每天在長大且伴隨疼痛。病患因此病症曾於前一年半內在其他醫院接受三次的切除手術；第一次為民

國 89 年 4 月 12 日，其病理診斷報告為下顎骨纖維性發育不良，但於十一個月後下顎骨仍出現相似腫塊；病患於民國 90 年 3 月 14 日在同一家醫院接受第二次之切除手術，但術後病理診斷報告為下顎骨骨性肉瘤。醫師曾於術後向病患建議需再進一步施行下顎骨廣泛切除術、頸部淋巴結分離術、及腓骨游離皮辦重建手術，但病患當時並未接受醫師之建議，也未接受化學治療。三個月後腫瘤又再度復發，但病患僅願意接受切除手術。其病理診斷報告為下顎骨纖維骨性病灶。

由於下顎骨疼痛且腫瘤漸漸變大，患者前來本院求診，理學檢查發現整個下顎骨從右側骨角到左側骨體有一個如石頭般硬度、大約 10 cm×6 cm×6 cm 之腫塊(圖一～四)，身體其他部位並未發現其他病灶。病患家族成員中並無罹患相關之疾病，也無相關之家族病史。其環口 X 光片(圖五)發現下顎骨從右側骨角到左側骨體有陽光射線影像(Sun ray appearance)，電腦斷層影像中發現下顎骨從右側骨角到左側骨體都被腫瘤侵犯並於聯合部位(Symphysis)處的頰側及舌側穿出下顎骨，兩側頸部有小的淋巴結發現(圖六)[19-21]。胸部 X 光片、腹部超音波、及全身核子醫學骨頭掃描(圖七)等檢查並未發現有轉移之病灶，其他的生化檢驗報告皆在正常範圍內。於是於民國 91 年 10 月 8 日施行兩側肩胛舌骨肌上淋巴結分離術(Supraomohyoid lymph node dissection)、塊狀下顎骨切除術、口內腫瘤廣泛切除術、及小腿腓骨骨頭皮膚游離皮瓣(Free fibula osteocutaneous flap)重建手術，並安排術後每月一次，共六次療程之化學治療。重建手術是由本院整形外科完成(圖八～十)。

病患術後恢復順利，而且病理報告診斷為下顎骨高度骨性肉瘤，腫瘤穿出下顎骨皮質骨並侵犯軟組織，手術邊緣無殘留腫瘤組織，兩側頸部淋巴結也無腫瘤之轉移(圖十一～十二)。病患於民國 91 年 10 月 29 日出院，並接受門診追蹤治療(圖十三～十四)。

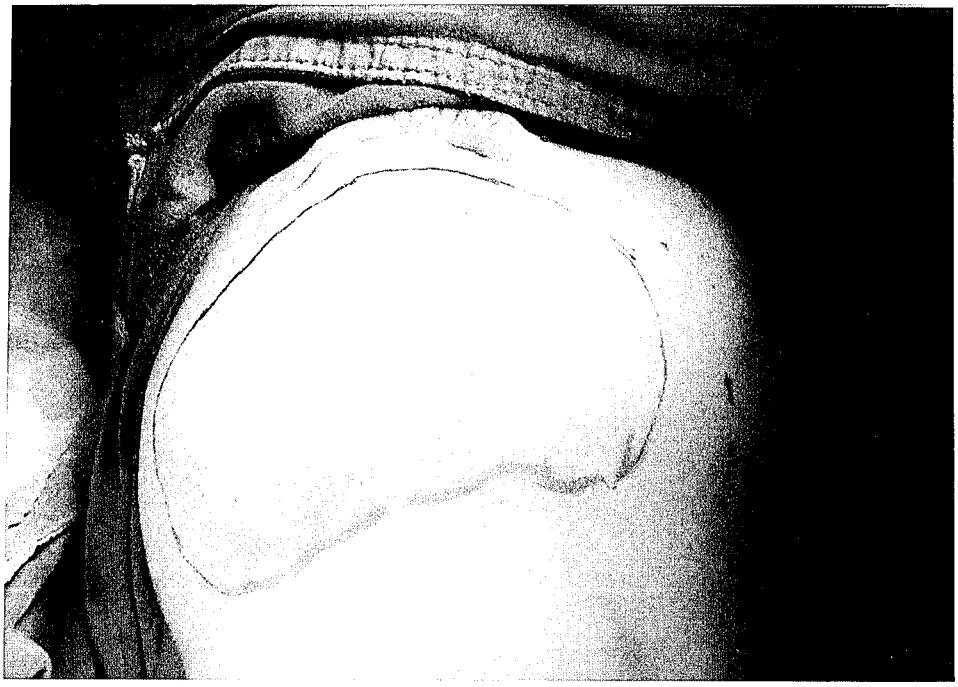
病患於民國 91 年 11 月 25 日開始進行每月一次共六次之骨性肉瘤之化學治療[22-28]，使用之藥物為 Dacarbazine 420 mg (300 mg/m^2)，Epirubicin 28 mg (20 mg/m^2)，Ifosfamide 3000 mg (2500 mg/m^2)，Mensa 3000 mg (2500 mg/m^2)。病患可接受完六次之化學治療療程，並規律的接受門診追蹤，目前術後追蹤七個月，下顎骨並無腫瘤復發之清況。

討　論

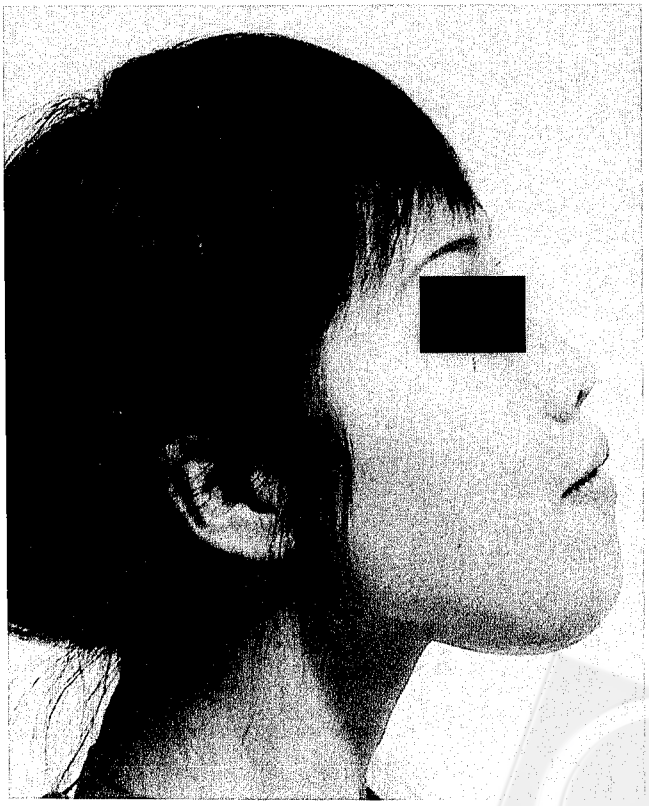
本篇報告是一罕見的下顎骨骨性肉瘤之病例，因下顎骨骨性肉瘤相當罕見，並未有大規模病例數之相關統計，及不同治療方式對其癒後影響之相關研究[29-36]。主要之文獻為頭頸部骨性肉瘤之綜合報告[7, 15, 18, 38-42]，其內容整理如表一。本篇提出此一病例之病患病史、理學檢查、放射線檢查、核子醫學檢查、檢驗報告、診斷、治療方式、及癒後，作為日後類似病例研究之參考。

骨性肉瘤發生的原因仍不明，有學者認為造成的因素可能有三個[43]：1).先前的良性骨疾，如 Paget 氏病與纖維性病變；2).放射線；3).外傷。Yannopoulos 也報告過在顏面骨纖維性發育不良病引起之骨性肉瘤[44]。此篇報告與本病例之病程有相類似之處，於發病初期曾被診斷為下顎骨纖維性發育不良。但本病例之腫瘤復發速度快，由纖維性發育不良病引起的骨性肉瘤之病程較慢[37]，這一點可以提供大家日後遇到類似的骨骼病灶時，需將骨性肉瘤列入鑑別診斷。

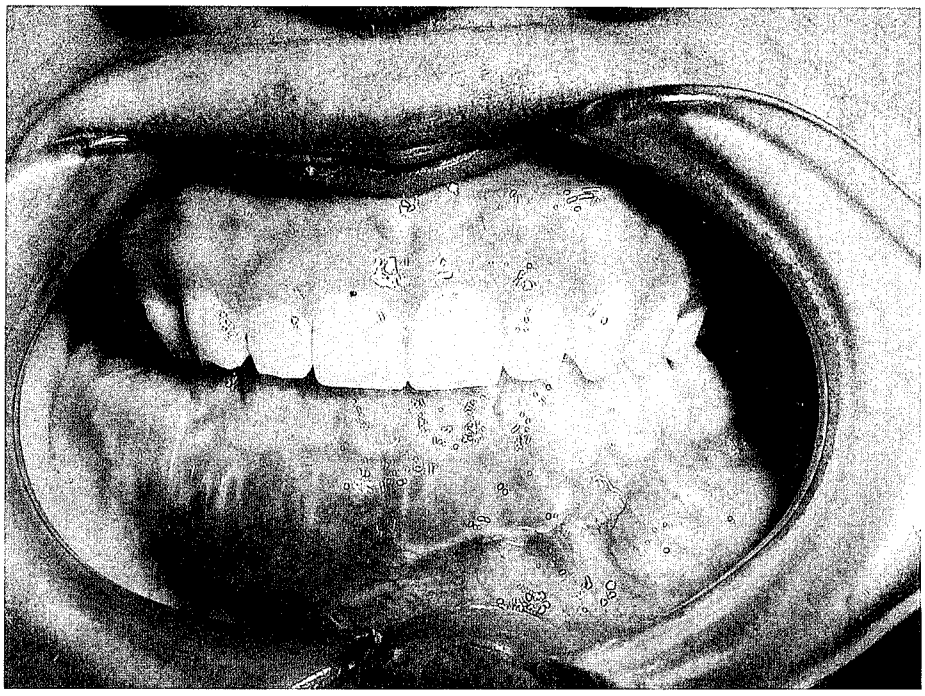
下顎骨骨性肉瘤之診斷，在 X 光片上，可呈現放射線可透射性、不透射性或兩者均有；有時在早期並無骨破壞跡象可見[45]；有時可見陽光射線影像(Sun ray appearance)，但此現象亦可見於其他疾病，如骨髓瘤、轉移癌、Ewing 氏肉瘤、結核病及其他炎性骨疾。陽光射線影像呈現於顎骨的骨性肉瘤者只有 25%[46]。也有報告認為牙周韌帶空間變寬也可作為早期診斷之依據[47]，因為鄰近病灶區的齒槽骨對腫瘤侵犯較無阻力。但此現象亦可見於其他疾病，如軟骨



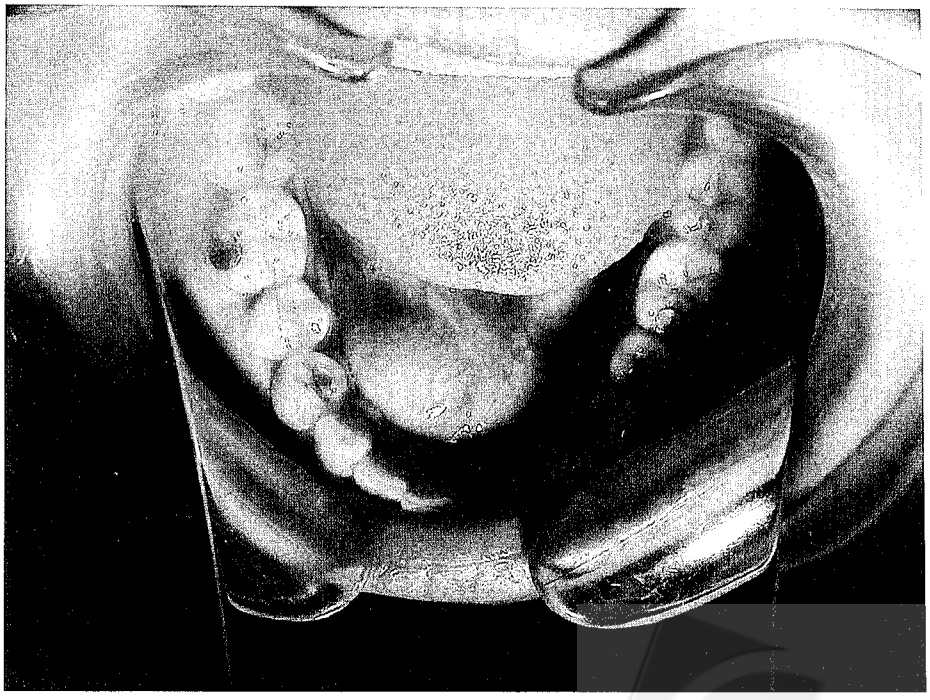
圖一　正面下方觀之臨床病灶。



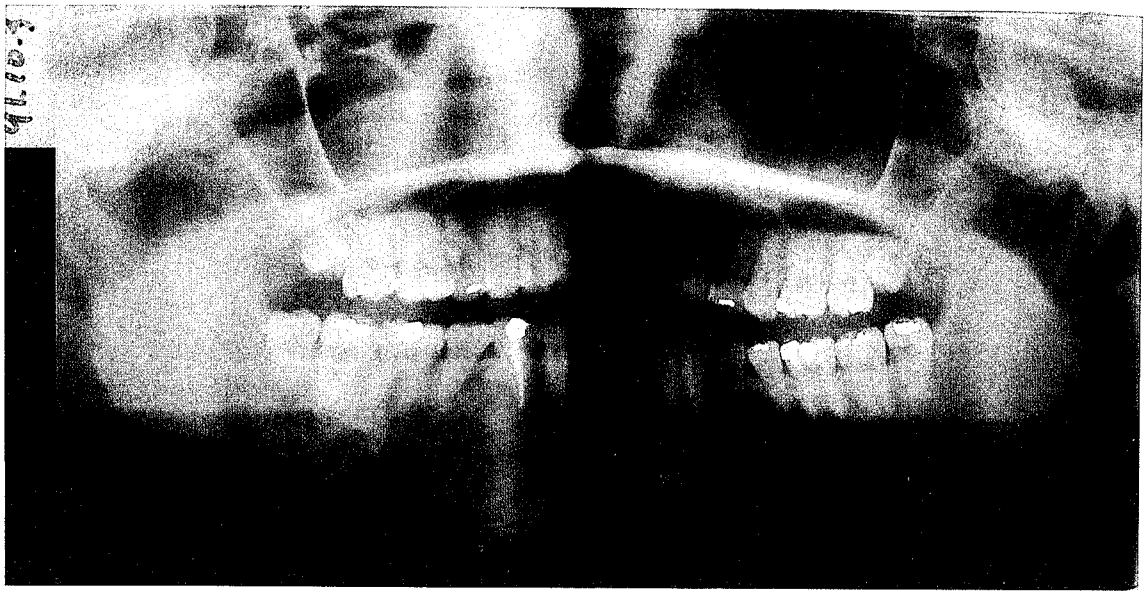
圖二　側面觀之臨床病灶。



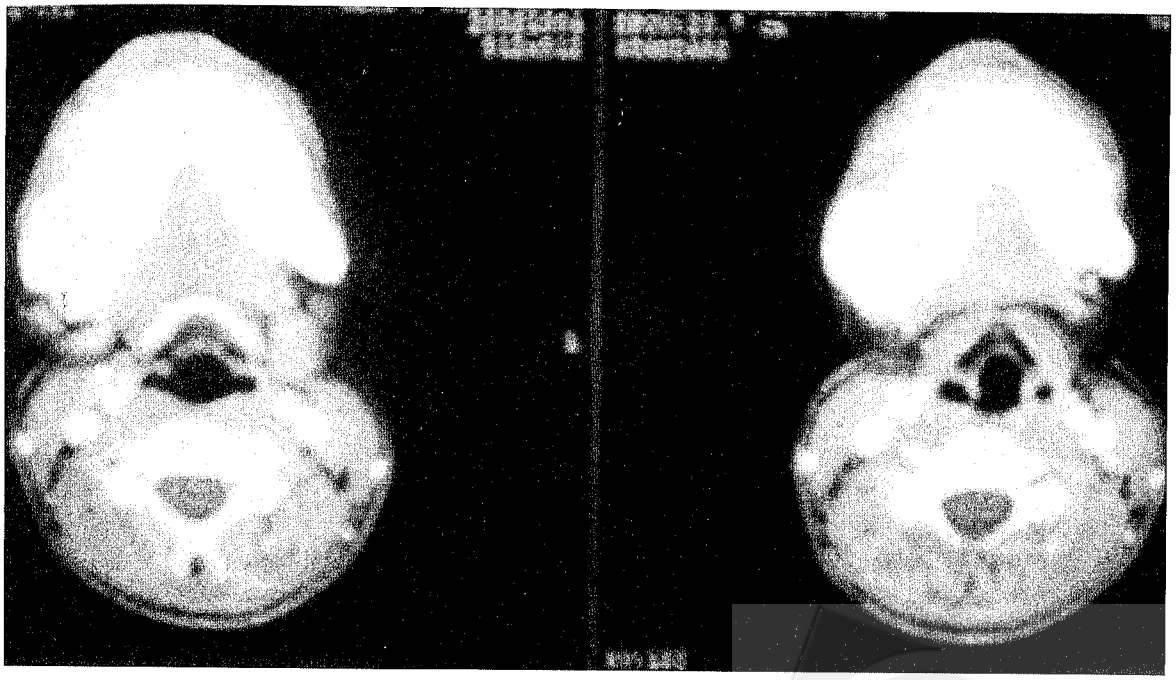
圖三　口內正面觀之臨床病灶。



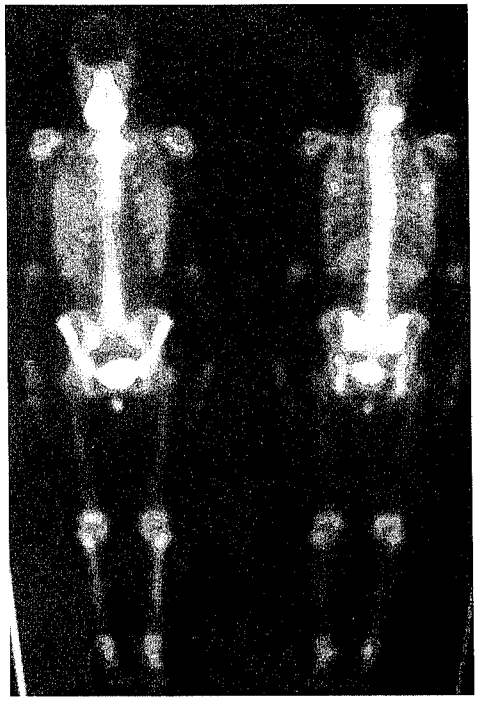
圖四　口內上面觀之臨床病灶。



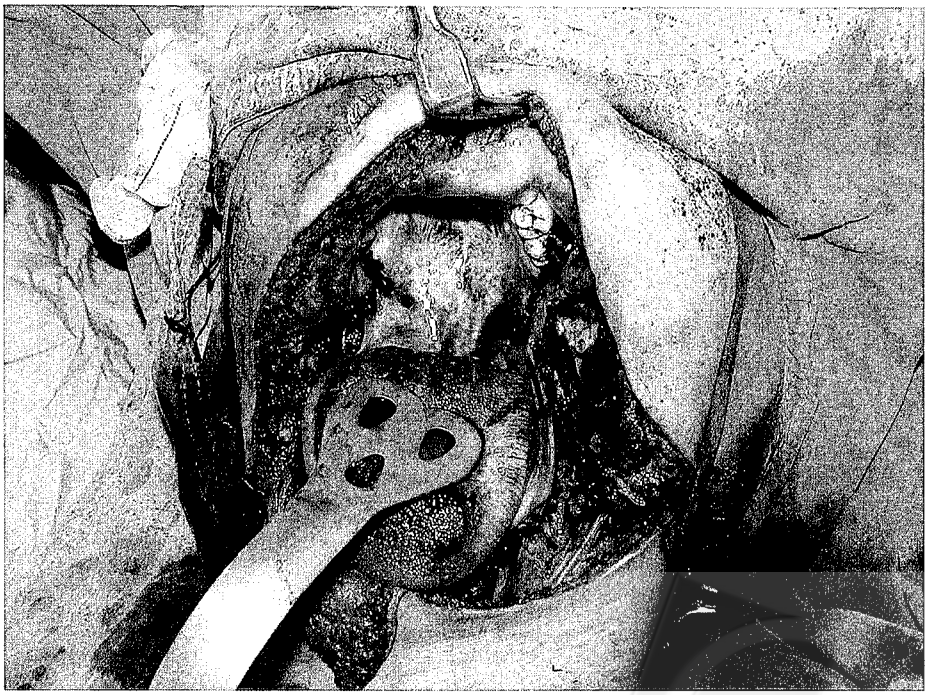
圖五　環口 X 光片。



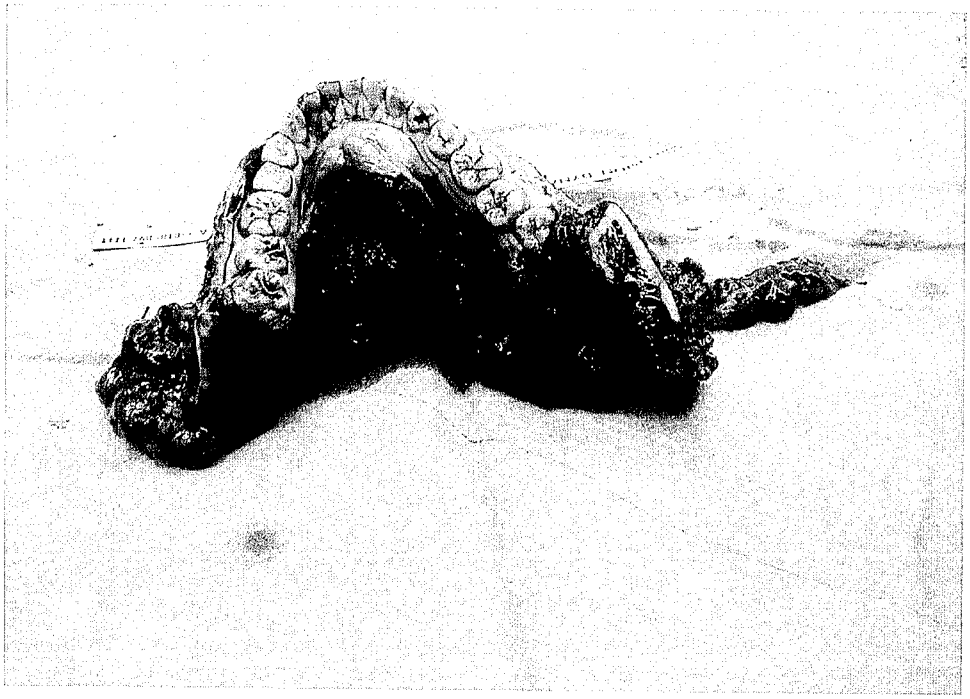
圖六　電腦斷層影像。



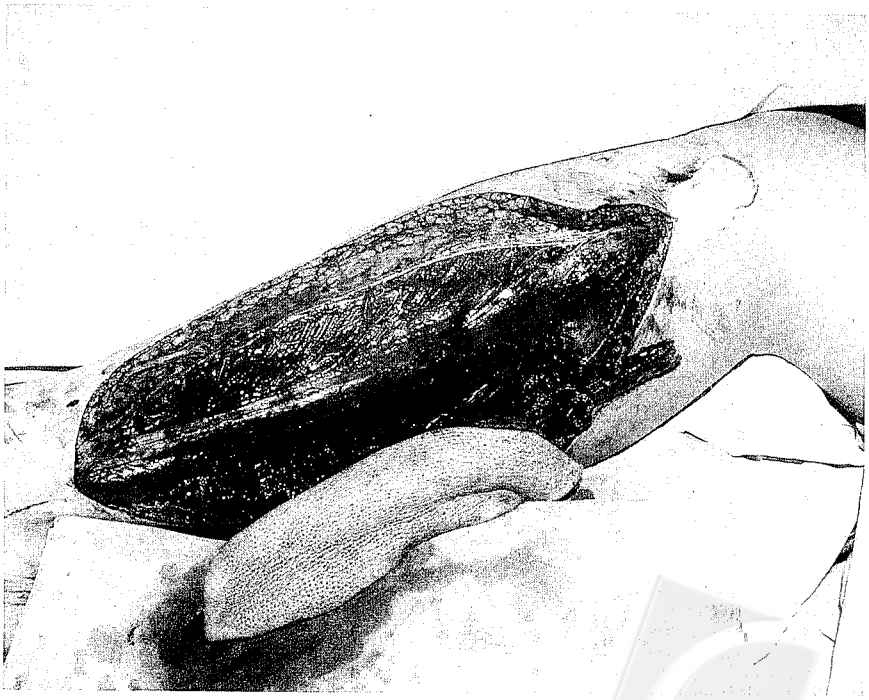
圖七　全身核子醫學骨頭掃描。



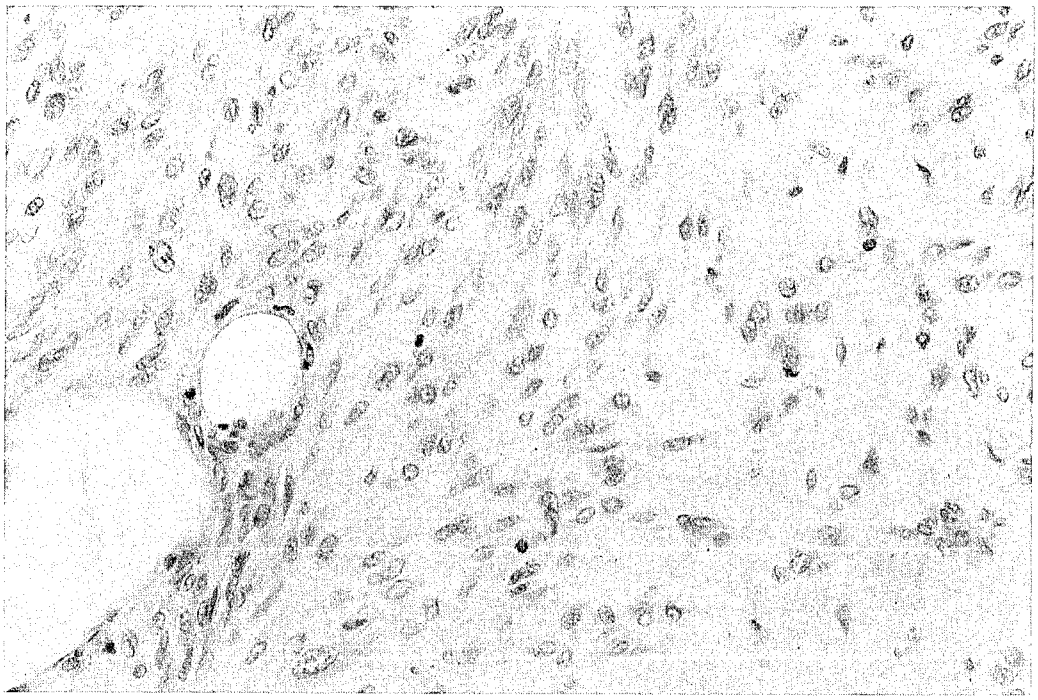
圖八　手術切除後正面觀。



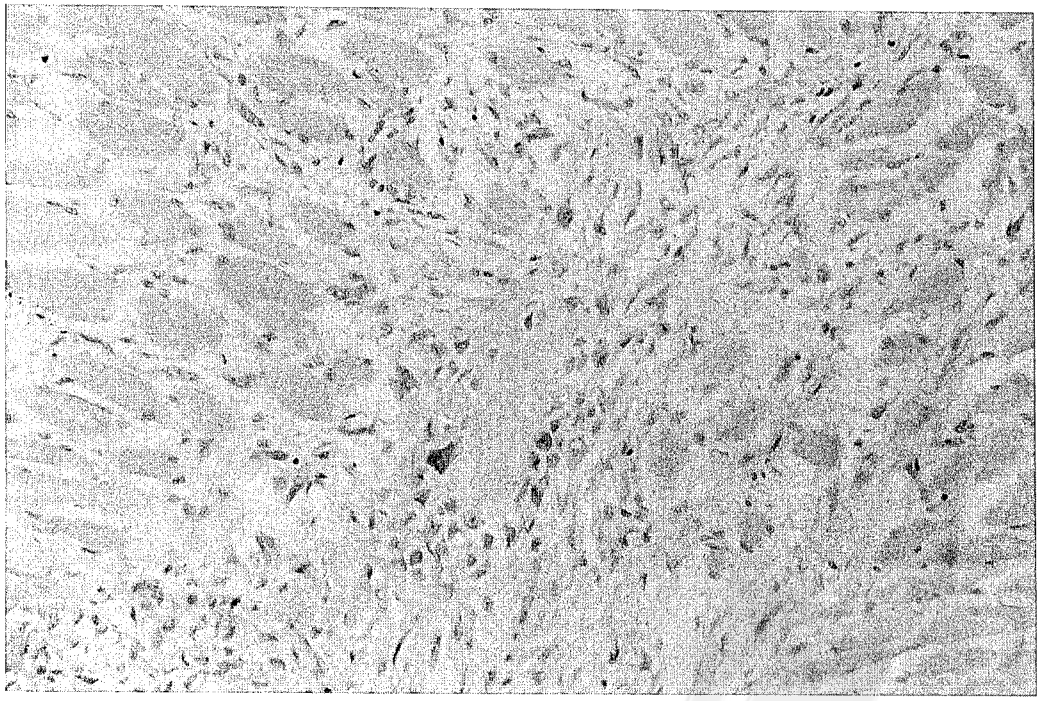
圖九　腫瘤標本。



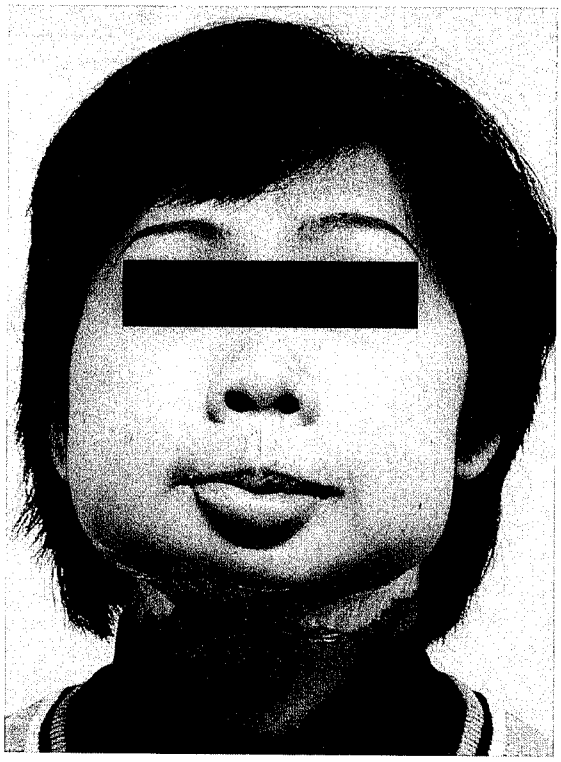
圖十　小腿腓骨骨頭皮膚游離皮瓣。



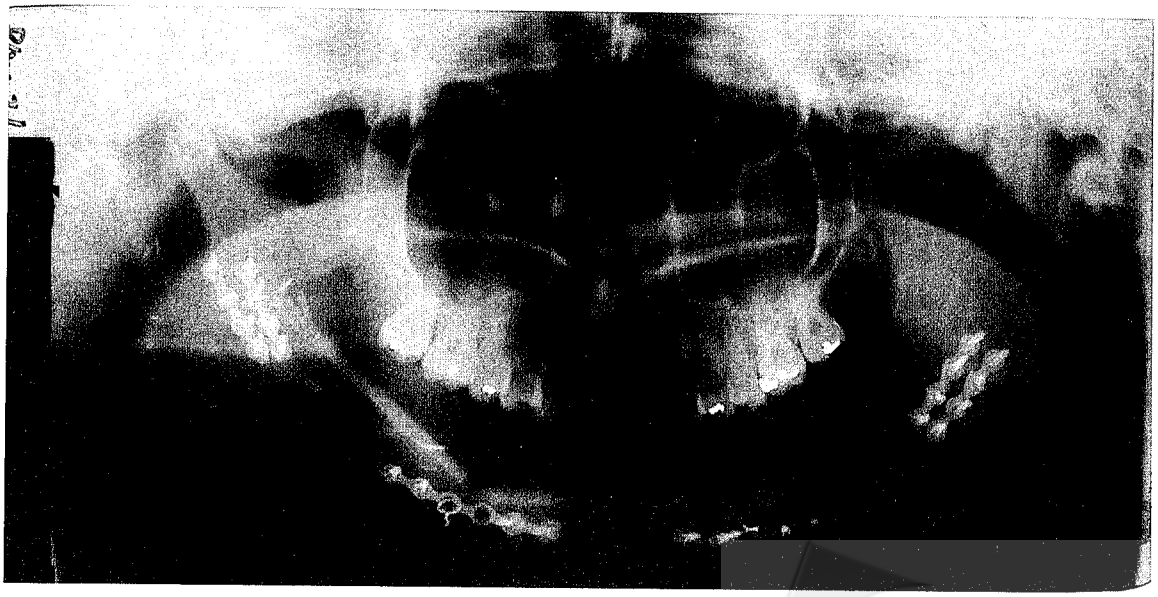
圖十一　200 倍下 H&E stain 之腫瘤細胞。



圖十二　100 倍下 H&E stain 之腫瘤骨質部分。



圖十三　手術後正面觀。



圖十四　手術後環口 X 光片。

表一 頭頸部骨性肉瘤之文獻報告。(S:surgery; R:radiotherapy; C:Chemotherapy; NG:non given)

作者機構國家	病例收集年數	病例數	每年平均病例數	病患平均年齡	男性比女性	腫瘤部位	治療方式	整體存活率
Oda *et al*,[15] Univ. of Washington, Seattle, WA	18 (1981-1996)	13	0.86	40.9	1.6:1	46% mandible; 23% maxilla; 15% orbit; 16% craniofacial	S; R; S + C; S + C + R	72% (5 yr)
Delgado, *et al*,[38] Instituto Nacional de Cancerologia, Mexico City, Mexico	18 (1972-1990)	23	1.27	28 (maxilla, 25 yr; mandible, 38 yr)	0.8:1	52% maxilla 48% mandible	S, 91% R, 61% C, 48%	10% (5 yr)
Wanebo *et al*,[39] Society of Head & Neck Surgeons: Head & Neck Sarcoma Registry, USA	8 (1982-1990)	29	3.62	NG	NG	NG	S; S + R; S + C; S + C + R	45% (5 yr)
Salvati *et al*,[40] La Spaienza, Rome, Italy	36 (1953-1989)	19	0.53	34.6	0.5:1	100% skull, craniofacial	S ± R S + C S + R + C	10% (2 yr)
Bertoni *et al*,[41] Instituto Rizzoli, Bologna, Italy	37 (1950-1987)	28	0.76	36.9	1.15:1	71% mandible 29% maxilla	S ± R	23% (5yr)
Mark *et al*,[7] UCLA, Los Angles, CA	32 (1955-1987)	18	0.56	28	1.0:1	50% mandible 33% maxilla 11% skull 5% orbit	S; R; C; S + R S + C S + R + C C + R	47% (5 yr)
Goepfert *et al*,[42] M.D. Anderson Cancer Center, Huston, TX	34 (1953-1987)	70	2.05	32	1.3:1	40% mandible 38% maxilla 21% craniofacial	S; R; C; S + R; S + C; C + R; R + C	53% (2 yr) 40% (5 yr)
Chao WY *et al*,[18] Taiwan Univ. H, Taiwan	17 (1965-1981)	5	0.29	31.4	1.5:1	20% mandible 60% maxilla 20% craniofacial	S; S + R;	80% (1.6 yr)

肉瘤及硬皮病等。骨性肉瘤確實之診斷還是需要組織切片來證實，因此取得足夠且具代表性之組織標本，方能確立診斷。

下顎骨骨性肉瘤之治療，以廣泛切除為主。但頭頸部之骨性肉瘤，無法像長骨部位的病灶可以廣泛切除，此病灶復發率很高。手術切除後，除了局部復發外，遠端轉移很常見，多半在二年內發生，尤其肺部轉移佔 90%[48]。因此其治療方式除了手術及局部放射線治療外，也有學者建議加上預防性肺部放射線治療[49]及化學治療。另外也有報告建議可以施行手術前放射線治療、化學治療[50]或經由動脈之化學治療(Intra-arterial chemotherapy)；由於目前並無下顎骨骨性肉瘤之治療與癒後等廣泛性相關研究，因此其治療方式沒有固定式準則；目前治療此類病患，大多參考身體其他長骨部位之骨性肉瘤之治療準則[51-55]。由於下顎骨為一橫跨身體左右兩側之骨頭，當下顎骨骨性肉瘤發生或已經影響左右兩邊時，其治療時需考慮兩側轉移的問題；使用經由動脈之化學治療時，也須注意到身體左右兩側同時進行之高風險；這與身體其他長骨部位有相同病症的處理有很大的不同[56-60]。

下顎骨骨性肉瘤之癒後，依其原發腫瘤大小，手術切除邊界是否侵犯，有無接受術前、術後化學治療或放射線治療而有所不同；整體存活率依研究不同而有不同的結果(如表一)。本病例接受廣泛性手術切除及術後化學治療七個月之後，仍沒有復發的現象，但仍需密切的追蹤以觀察治療之成效。

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Osteogenic Sarcoma of the Mandible - A Case Report

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Abstract

Osteogenic sarcoma is a rare disease in the head and neck region. A case of osteogenic sarcoma of the mandible was reported. The 25 year-old female visited our OPD with chief complain of stony hard swelling of whole mandible. In the past one and half years, the patient ever received three operations of this lesion. The pathological reported of these three operations were fibrous dysplasia of mandible, osteogenic sarcoma of mandible and fibrous-osseous lesion of mandible. After physical examination and imaging examination, the osteogenic sarcoma of the bilateral mandible body, angle and symphysis was impressed. The patient received bilateral supraomohyoid neck lymph node dissection, block mandibulectomy, wide excision of tumor, and free fibula osteo-cutaneous flap reconstruction. The patient also received adjuvant chemotherapy after two months after the operation. The osteogenic sarcoma of mandible is a rare disease, so reported this patient.

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