Long-Term Outcomes of Surgical Management in the Patients with Facial Congenital Melanocytic Nevi

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Background*: The challenges of management of facial congenital melanocytic nevi (CMN) are the balance of the risk of malignant transformation, surgical management and the long-term evaluation of the functional and cosmetic outcomes.*
Objective*: To present information on the long-term surgical management outcomes of patients with facial CMN, which may be applicable for future clinical and surgical approaches for these lesions.*
Material and Method*: A follow-up study was performed of patients with facial CMN that were clinically evaluated by the author at Srinagarind Hospital, Khon Kaen University, between 1993 and 2011. The clinical records, photographs, surgical managements and outcomes were analyzed.*
Results*: Among the 20 patients assessed, the female-to-male ratio was 2.33 to 1 with one patient being a twin. Most (95%) had a single and ten a large CMN. The surgical treatments of these lesions included: serial excision, excision with full thickness skin grafts, excision with composite graft, excision with local and regional flaps, and excision with tissue expanders and flap. The long-term outcome revealed that no patient's condition changed or developed into melanoma. Most of the patients had an acceptable cosmetic and functional outcome. One patient had a hypertrophic scar on the face, one a mild ectropion of the lower eyelid and 2 hyperpigmentation of the skin-grafted area.*
Conclusion*: The author presents a number of surgical techniques that may be used for decision-making in surgical management of each CMN. For the most part, analysis of the characteristics of the CMN, comprehensive evaluation of the anatomic composition of the defects and application of good reconstructive methods will provide acceptable long-term surgical outcomes and reduce the psychological impact to parents and patients. Early surgical removal of large CMNs is recommended and long-term follow-up until adulthood are the two essentials.*

Keywords*: Facial congenital melanocytic nevi (CMNs), Malignant potential, Surgical management and reconstruction, Long-term outcome*

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Congenital melanocytic nevi (CMN) are melanocytic nevi with distinct histopathologic features and may be present at birth or within the first year of life. The size of the lesions may vary considerably, from a few millimeters to several centimeters, covering wide areas in different anatomic locations. Many authors classify CMN according to their size and the greatest diameter into small (diameter < 1.5 cm), medium (diameter 1.5 to 20 cm), large (diameter >20 cm) and giant (diameter > 50 cm)[1]. The large CMN-defined as > 20 cm in greatest dimension in adulthood-corresponds to a 9-cm scalp or a 6-cm trunk lesion in an infant[2] or about 2% or more of body surface. The classification is helpful in both prediction of prognosis and challenges in reconstruction. The most common anatomic location for a large CMN is the trunk, followed by the legs, arms, and head and neck[3]. The differential diagnoses for CMN include epidermal nevus, nevus sebaceous, cafe-au-lait spot, and Mongolian spot.

The incidence of CMN among newborns ranges between 0.2% and 6%[4-6]. The incidence of large/giant CMN is rare (0.0005% or 1 in 20,000 live births)[6,7]. An accurate estimation of the risk of developing melanoma in persisting CMN are difficult to obtain[7] and is believed to be proportional to the size of the CMN. The lifetime risk for melanoma arising in small CMN is not well established and is perhaps between 0% and 5% and the risk of giant CMN between 5% and 10%[8]. Schaffer suggested that the incidence ranges for giant CMN is between 5% and 10% over a lifetime[9] while the Danish Birth Registry estimated a lifetime risk of approximately 6.3%[10] and DeDavid et

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al[11] found 12% of giant CMN developed into melanoma within their lesions. Although it is extremely rare to find a melanoma present at birth, or in infancy; most cases were found to occur within the first decade of life[12,13].

The challenges for management of CMN, especially of the facial area, are the balanced goals of CMN removal (or decreasing the risk of the developing melanoma) and the restoration of function and near-normal appearance after surgical removal.

The objectives of the present study were to (a) present the treatment methods (b) surgical managements and (c) long-term outcomes of the patients with facial CMN at Srinagarind Hospital, Khon Kaen University, between 1993 and 2011. The analysis results of these data could be useful for making recommendations for appropriate guidelines in diagnosis and treatment of these patients. The study is among the first reports on long-term surgical outcomes of facial CMN in Thailand; the information documents the current state of knowledge and is therefore applicable for setting clinical and surgical approaches to facial CMN.

Material and Method

Setting

Srinagarind Hospital is a university hospital and a major tertiary referral center in Khon Kaen, a province in the heart of the Northeast Thailand, a geopolitical region with a population of ~22 million.

Study Design

The medical records of the patients with facial CMN were reviewed, managed by, and/or with consultation by the author. The diagnosis of CMN was accomplished through clinical and/or pathologic reports. The details of different and serial reconstructive surgeries as well as the final cosmetic, functional outcome and the patient's and his/her family's satisfaction were analyzed.

The protocol of the present study was reviewed and approved by the Ethics Committee of Khon Kaen University, based on the Helsinki Declaration, and written informed consent for adults (or assent for minors) was obtained from each patient.

Results

Between 1993 and 2011, 20 patients with facial CMN were analyzed. The female-to male ratio was 2.33 to 1 (14 girls and 6 boys) with one patient who was a twin. The majority of patients had a single CMN (19 patients, 95%) and one patient (5%) had two CMNs. Ten patients had CMNs classified as 'large' and 10 had medium-sized CMNs. Table 1 presents the demographic details and Fig. 1 presents the geographic distribution of these 20 patients with facial CMN treated at Srinagarind Hospital, Khon Kaen University, between 1993 and 2011. The highest incidence recorded was in Khon Kaen and Kalasin Provinces.

The surgical treatments of these lesions are serial excision, excision with full thickness skin grafts, excision with composite graft, excision with local flaps, and excision with tissue expanders and flaps. Patient No. 12 did not receive treatment and was lost to follow-up. The long-term outcome of the other 19 patients-according to an analysis of their medical records-was performed and revealed that none of them developed melanoma and all of them were satisfied with the treatment. Most of the patients had good cosmetic and functional outcomes; albeit one patient had a hypertrophic scar on the face (patient No. 13), one had mild ectropion of the lower eyelid (patient No. 9) and two had hyperpigmentation of the skin-grafted area (patients No. 5 and 15).

Selected Patient Reports

Patient No. 4

A male, twin, born in 1990 in Khon Kaen province, presented with a large hairy nevus on the forehead, left temporal area and left eyebrow. Staged excision with tissue expansion was performed in 1996 and a composite hair baring skin graft to the left eyebrow was performed in 2001. On follow-up in 2011, at the age of 21 years, his family and he were satisfied with his facial appearance.

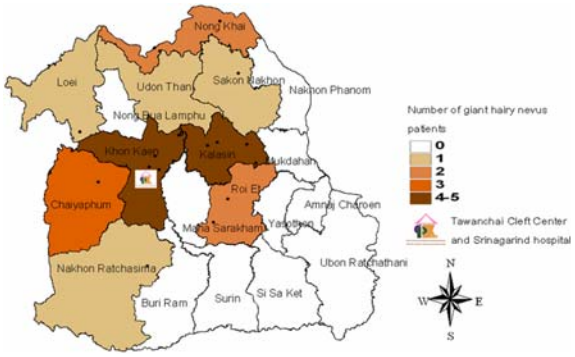

Fig. 1 Geographic distribution of patients with facial CMN treated at Srinagarind Hospital between 1993 and 2011

Table 1. Details of the 20 patients with facial CMN treated at Srinagarind Hospital, Khon Kaen University between 1993 and 2011

Patient No.	Year of birth	Province	Diagnosis and involved area	Treatment
1	1968	Kalasin	Large CMN; right cheek, temporal area	Excision with tissue expansion
2	1990	Khon Kaen	Medium CMN; upper lip	Excision with full thickness skin graft and local flap
3	1992	Khon Kaen	Medium CMN; right oral commissure	Serial excision
4	1990	Khon Kaen	Large CMN; forehead, left temporal area and left eye brow	Staged excision with tissue expansion and composite hair baring skin graft to left eyebrow
5	1992	Chaiyaphum	Large CMN; left cheek	Excision with full thickness skin graft
6	1987	Nong Khai	Medium CMN; left nasolabial fold	Serial excision
7	1995	Khon Kaen	Medium CMN; right upper eyelid	Excision with full thickness skin graft
8	1993	Roi Et	Large CMN; left cheek, temporal area, left medial canthal area and nasal dorsum	Staged excision with cervicofacial flap and full thickness skin graft from post auricular area to temporal area and nasal dorsum
9	1993	Chaiyaphum	Large CMN; left cheek and nasal dorsum	Excision with cervicofacial flap and full thickness skin graft
10	1964	Nong Khai	Medium CMN; lateral canthus involve upper and lower eyelid	Excision with lid switch flap and rotation cheek flap
11	1987	Loei	Medium CMN; upper lip	Excision with full thickness skin graft
12	1997	Udon Thani	Large CMN; right cheek	-
13	1996	Kalasin	Giant CMN; right cheek, nose, bilateral canthal area, medial part of right upper eyelid	Excision and staged reconstruction with cervicofacial flap and full thickness skin graft; scar revision and lateral canthopexy
14	1999	Khon Kaen	Large CMN; left cheek, involving lateral pert of nose	Excision with cervicofacial flap and full thickness skin graft
15	1997	Kalasin	Medium CMN; Tip of the nose, alar area, philtrum and upper lip	Excision with full thickness skin graft and subsequent dermabrasion
16	1992	Roi Et	Compound nevus; left median canthus	Excision with full thickness skin graft
17	2000	Nakhon Ratchasima	Large CMN; left cheek, nasal dorsum and left leg	Excision and split thickness skin graft to left leg
18	1998	Chaiyaphum	Large CMN; left forehead and face	Excision and tissue expansion
19	1996	Sakon Nakhon	Medium CMN; upper lip	Excision with full thickness skin graft
20	2002	Kalasin	Medium CMN, Left medial canthal area	Excision with local flap

Patient No. 5

A female patient, born in 1992 in Chaiyaphum province, presented with a large hairy nevus on the left cheek. Excision with full thickness from the buttock area was performed in 1995. On follow-up in 2011, at the age of 19 years, she had mild pigmentation of the skin-grafted area.

Patient No. 8

A female patient, born in 1993 in Roi Et

Fig. 2 Pre- and post-operative pictures (including at follow-up at age 18) of a male patient and his twin. He presented with a large hairy nevus at the forehead, left temporal area and left eyebrow. It was treated by staged excision with tissue expansion and composite hair baring skin graft to left eyebrow

province, presented with a large hairy nevus at the left cheek, temporal area, left medial canthal area and nasal dorsum. Staged excision with cervicofacial flap and full thickness skin graft-from the post auricular area to the temporal area and the nasal dorsum-was performed in 1995. On follow-up in 2011 (at the age of 18 years), her family and she were satisfied with her facial appearance.

Patient No. 13

A male patient, born in 1999 in Khon Kaen province, presented with a large hairy nevus involving the left cheek, nose, bilateral canthal area, and medial part of the right upper eyelid. Excision and staged reconstruction with a cervicofacial flap and a full thickness skin graft from the post auricular area was performed in 1998. A subsequent scar revision and lateral canthopexy was performed to correct ectropion on the right lower eyelid. On follow-up in 2011 (at 12 years of age), there was a good skin color match; however, there was also a hypertrophic scar at the scar edge on the right cheek. Minor scar revision was planned.

Patient No. 14

A male patient, born in 1999 in Khon Kaen province, presented with a large hairy nevus at the left

Fig. 3 Pre- and post-operative photos (at the age of 19 years) of a female patient who presented with a large hairy nevus at the left cheek, treated by excision with full thickness from the buttock

Fig. 4 Pre- and post-operative photos (including at follow-up at 18 years of age) of a female patient who presented with a large hairy nevus at the left cheek, temporal area, left medial canthal area and nasal dorsum, treated by staged excision with cervicofacial flap and full thickness skin graft to temporal area and nasal

cheek, involving lateral part of nose. Excision with a cervicofacial flap and a full thickness skin graft from the post auricular area (to the temporal area) was performed in 2000. On follow-up in 2011 (at the age of 12 years), there was a good skin-color match on the left side of the face. Minor scar revision was planned.

Patient No. 15

A male patient, born in 1997 in Kalasin province, presented with a medium hairy nevus involving the tip of the nose, alar area, philtrum and the upper lip. Excision with a full thickness skin graft from the post-auricular area was performed in 2001. Dermabrasion was performed in 2011. On follow-up in

Fig. 5 Pre- and post-operative photos (including at follow-up at age 12) of a male patient who presented with a large hairy nevus involving the left cheek, nose, bilateral canthal area, and medial part of the right upper eyelid. He was treated by excision and staged reconstruction with a cervicofacial flap and a full thickness skin graft and a subsequent scar revision and lateral canthopexy

Fig. 6 Pre- and post-operative photos (including at follow-up at age 12 years) of a male patient who presented with a large hairy nevus on the left cheek, involving lateral part of the nose, treated by excision with a cervicofacial flap and a full thickness skin graft from the post auricular area

2011 (at age 14 years) he and his family were satisfied with the results, even though there was mild hyperpigmentation of the skin-grafted area.

Fig. 7 Pre- and post-operative photos of a male patient, who presented with a hairy nevus involving the tip of the nose, alar, philtrum and upper lip, treated by excision with a full thickness skin graft from the post-auricular area. Dermabrasion was performed at a later date

Patient No. 16

A female patient, born in 1994 in Roi Et province, presented with pigmented lesion at the left medial canthal area. Excision with a full thickness skin graft was performed in 2001. The pathology report indicated compound nevus. Minor scar revision was performed subsequently. On follow-up in 2011 (at 17 years of age), she and her family were satisfied with the results.

Patient No. 19

A female patient, born in 1996 in Sakon Nakorn province, presented with a hairy nevus at the upper lip. Excision with a full thickness skin graft was performed in 2005. On follow-up in 2011, at the age of 15 years, she and her family were satisfied with the results: there was good a skin-graft color-match. A minor scar revision was planned.

Discussion

Congenital melanocytic nevi (CMN) may appear as plaques, raised from the surrounding skin, with increased skin marking and/or appendages, and ranging in color from tan to deep blue-black. Natural

Fig. 8 Pre-operative photos and at follow-up (at age 17) of a female patient, who presented with pigmented lesion at the left medial canthal area, treated by excision with full thickness skin graft

Fig. 9 Pre- and post-operative photos (including at follow-up at the age of 15) of a female patient, who presented with a hairy nevus at the upper lip, treated by excision with a full thickness skin graft

elements of rugose texture or coarse terminal hair may be present[1]. The clinical findings of CMN may be round or oval; smooth, regular, and sharply demarcated and usually with a uniform pigmented pattern[14]. Changes may occur within the CMN that may concern clinicians, encouraging a decision for early surgical management; these include, a rupture of a hair follicle or a cyst, trauma, infection, changes to a dermatofibroma, seborrheic keratosis-like lesion[15], or even a malignant transformation which may present as an increasingly dark pigmentation, accelerated growth, a change in shape, the appearance of nodularity, pain, ulceration with or without bleeding, or pruritus.

The report lifetime risk of melanoma for small and medium CMN is 0 to 4.9% and typically post-pubertal[16]. Prophylactic excision of small and medium CMN can be advised anytime before puberty, in particular if it presents irritation or it is located in an area where any changes cannot be easily observed[1]. Watt et al (2007) conducted a systematic review of 8 studies, including a total of 432 giant CMNs, found patients had an increased risk of melanoma compared to the general population[17]. The management decision for large, medium and small CMNs must be done on a patient by patient basis and balance the risk of transition to melanoma and cosmetic and functional considerations[15]. Moreover, the indications for surgical excision of large/giant CMNs, other than to prevent melanoma, may include chronic pruritus, ulceration and infection[15].

The goal of surgical treatment is to remove as much of the CMN as possible while maintaining an acceptable level of function and achieving a satisfactory cosmetic outcome. Melanoma may arise in large/giant CMNs even in the first several years of life and surgical excision should be considered as early as possible. Excision of facial large CMN presents a distinct challenge to the plastic surgeon because of the concern of the risk of malignant degeneration and the psychologically damaging impact, to both the parents and the child, due to the unsightly lesions[18].

Many methods have been introduced for removal and reconstruction of large CMNs. Some methods have been introduced previously; including dermabrasion, chemical peels and lasers; but these are not effective for removing the nevus cells. Several surgical techniques have been described for management of large and giant CMNs[19,20]; including (a) a single stage, complete excision (including the deep margin) with primary closure and (b) a staged or serial excision, with tissue expansion, skin grafts or skin flaps[21]. A combination of these techniques is also often recommended and employed.

Of the 20 large and medium CMNs reviewed, the author's use of (a) tissue expanders for expanded flaps was performed on 3 patients (patients No. 1, 4 and 18) (b) a full-thickness skin graft in 11 patients, and (c) serial excision in 2 patients (patients No. 3 and 6). The management options were considered relative to pigmentation, malignant potential, and anatomical location of the respective lesions[20].

Adequate surgical excision-including the deep margin-to fascial level is recommended. Planning for excising CMN lesions in children includes taking adequate margins, and meticulous plastic surgery technique. Serial excision for facial lesions is less applicable than lesions on the trunk or extremities because of the concerns of anatomical distortion and functional disturbance. However, the technique has been used in the medium-sized lesions at the nasolabial fold and oral commissure.

Good, long-term outcomes for full thickness skin grafts from the post-auricular area occurred in patients No. 2, 5, 7, 8, 9, 11, 13, 14, 15, 16 and 19. However, skin graft hyperpigmentation did occur in two (patients

No. 5 and 15). The donor site of patient No. 5 was from the buttock area which may be a less preferable donor site for use as a full thickness skin graft to the face. Patient No. 15 had a habit of feeding buffalo fully exposed to the sun. The use of a full thickness skin graft donor site from above the supraclavicular area is preferable for reconstruction of a facial skin defect. For a larger defect, the use of an expanded, full thickness skin graft may be appropriate. Sun protection and proper skin management for prevention of skin hyperpigmentation should be advised for high-risk patients.

Tissue expansion for large/facial giant CMNs is a helpful technique in expanding the flap to achieve more functional and aesthetic goals and may be used as an expanded skin flaps or as an expanded full-thickness skin graft[22]. Composite hair baring skin graft was used in patient No. 4 for eye brow reconstruction. Additionally, the technique of a local flap (Fig. 11), staged reconstruction or a combination of various techniques (Fig. 2, 4, 5, 6 and 10) may be used. A program for prevention of hypertrophic scarring should also be used especially in the high-risk patients.

Additionally, treatment by a multidisciplinary team should be introduced. A good balance of social function vis-a-vis the treatment method and outcome will lead to full cooperation between the patient, the family and surgeons. The plastic surgeon has to educate the family about the natural history of CMN, the need for its removal and the realistic expectations of scarring and surgical outcome. When excision is recommended, he must advise on the best timing with consideration of the natural history of CMN, the physical and social development of the child and especially his/her facial growth.

The establishment of The Tawanchai Foundation for cleft lip, cleft palate and craniofacial deformities has provided timely and crucial assistance for patients (especially in the realms of accessibility to hospital, follow-up and some adjunctive management programmes), in addition to that which is provided by the current healthcare system in Thailand. Analysis of the geographic distribution of medium and large CMNs among those treated at Srinagarind Hospital and the Center of Cleft Lip-Palate and Craniofacial deformities can be helpful for planning for Center's provision of care for patients with CMN.

Conclusion

The author has presented the experience of a number of surgical techniques that provided acceptable and good surgical outcomes, which may be recommended for surgical decision-making for the management of each CMN. The timing for excision and reconstruction of large, facial CMNs should begin early and within the first year of life. The surgical treatment includes adequate excision, and proper reconstructive techniques. The expanded full thickness skin graft and flap is a good choice to provide the skin with optimum quality and texture. The comprehensive evaluation, appropriate reconstructive method and ancillary and multidisciplinary treatment, and long-term follow-up are essential.

Fig. 10 Photos of patient no. 2 with CMN of the upper lip involving labial mucosa, successfully treated by excision with a full thickness skin graft and a V-Y mucosa advancement flap

Fig. 11 Photos of patient No. 20, with CMN at the left medial canthus, successfully treated by excision with glabella transposition flap

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Potential conflicts of interest

None.

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ผลลัพธ์ระยะยาวของการรักษาทางการผ่าตัดในผู้ป่วยที่มีไฝดำที่ใบหน้าแต่กำเนิด

บวรศิลป์ เชาวน์ชื่น

***ภูมิหลัง**: ความท้าทายของการรักษาไฝดำที่ใบหน้าแต่กำเนิดคือ ความสมดุลระหว่างความเสี่ยงต่อการเปลี่ยนเป็นมะเร็ง การรักษาทางการผ่าตัด และการประเมินระยะยาวของผลลัพธ์ในด้านหน้าที่การทำงานและความสวยงาม*

***วัตถุประสงค์**: เพื่อนำเสนอสารสนเทศของผลลัพธ์ของการรักษาทางการผ่าตัดในผู้ป่วยไฝดำที่ใบหน้าแต่กำเนิดซึ่งสามารถนำไปประยุกต์ใช้เป็นแนวทางทางการรักษาทางคลินิกและการผ่าตัดสำหรับรอยโรคเหล่านี้ในอนาคต*

***วัสดุและวิธีการ**: เป็นการติดตามผู้ป่วยไฝดำที่ใบหน้าแต่กำเนิดที่ได้รับการประเมินทางคลินิกและรักษาโดยผู้นิพนธ์ในโรงพยาบาลศรีนครินทร์ มหาวิทยาลัยขอนแก่น ระหว่างปี พ.ศ. 2536-2554 รวมถึงการวิเคราะห์บันทึกทางการแพทย์ การรักษาทางการผ่าตัด และผลลัพธ์*

***ผลการศึกษา**: ผู้ป่วย 20 ราย มีอัตราเพศหญิงต่อเพศชาย เป็น 2.33 ต่อ 1 ผู้ป่วย 1 ราย เป็นคู่แฝด ผู้ป่วยส่วนใหญ่ (ร้อยละ 95) มีรอยโรคแห่งเดียว และ 10 ราย เป็นชนิดขนาดใหญ่ การรักษาทางการผ่าตัดของรอยโรคเหล่านี้ประกอบด้วย การผ่าตัดแบบตัดหลายครั้ง การตัดออกร่วมกับการปลูกถ่ายผิวหนังแบบทั้งส่วนของความหนา การตัดออกร่วมกับการปลูกถ่ายเนื้อเยื่อผสม การตัดออกร่วมกับการใช้แผ่นเนื้อเฉพาะที่ และแผ่นเนื้อจากบริเวณข้างเคียง และการตัดออกร่วมกับการขยายเนื้อเยื่อ และการใช้แผ่นเนื้อ ผลลัพธ์ระยะยาวพบว่าผู้ป่วยทุกรายไม่มีการเปลี่ยนแปลงเป็นมะเร็งผิวหนังชนิดเมลาโนมา ผู้ป่วยส่วนใหญ่มีผลลัพธ์ด้านความสวยงาม และหน้าที่การทำงานเป็นที่พึงพอใจ ผู้ป่วย 1 ราย เกิดแผลเป็นนูนบนใบหน้า และอีก 1 ราย เกิดการดึงรั้งออกของเปลือกตาล่าง และอีก 2 รายเกิดสีคล้ำขึ้นของบริเวณที่ปลูกถ่ายผิวหนัง*

***สรุป**: ไฝดำที่ใบหน้าขนาดใหญ่และขนาดกลางมีความเสี่ยงที่มากกว่าในการเปลี่ยนแปลงเป็นมะเร็ง และมีความท้าทายในด้านการรักษาและการผ่าตัดเสริมสร้าง ผู้นิพนธ์นำเสนอวิธีการผ่าตัดชนิดต่างๆ ที่อาจนำไปใช้เป็นแนวทางการตัดสินใจของการผ่าตัดรักษาในผู้ป่วยที่มีไฝดำใบหน้าแต่ละราย การวิเคราะห์ลักษณะของไฝดำการประเมินทั้งส่วนประกอบและลักษณะกายวิภาคของความบกพร่อง และการประยุกต์วิธีการผ่าตัดเสริมสร้างที่ดีจะให้ผลลัพธ์ด้านการผ่าตัดในระยะยาวที่ดีและลดผลกระทบทางด้านจิตวิทยาสังคมต่อผู้ป่วยและผู้ปกครอง ผู้ป่วยที่มีไฝดำขนาดใหญ่ควรได้รับการตัดออกตั้งแต่ในระยะขวบปีแรกและควรมีการติดตามจนเป็นผู้ใหญ่*